

## REVIEW

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**LAGOCHILASCARIASIS: AN EMERGING  
PARASITIC DISEASE**

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**ABSTRACT**

Lagochilascariasis, described for the first time in 1909, is caused by a helminth nematode within the *Lagochilascaris* genus, and Ascarididae family, which mainly affects the underprivileged, especially among rural populations. Brazil presents more than 75% of the cases worldwide, mostly in the Amazon region, where the illness is considered emergent. The main clinical manifestation in humans is the appearance of subcutaneous purulent lesions normally found around the neck, mastoid and middle ear regions, the originating form is a painless granulomatous reaction that may have a chronic course and lead to death. Considering the paucity of reports on this helminthiasis, a bibliographic study was performed with the intent of consolidating information found systemized as etiology, epidemiology, pathogeny, clinical aspects, diagnostic, treatment, prophylaxis and control, as well as guiding health professionals in the resolution of human lagochilascariasis cases. The present review reinforces the need for further studies on the subject, with a view to increasing the appropriate management of the disease and learning from it, to improve control, based on adequate knowledge of its natural cycle, as well as health education of the population.

**KEY WORDS:** Emerging diseases; human lagochilascariasis; infectious diseases.

**INTRODUCTION**

Lagochilascariasis is a zoonosis caused by a helminth of the Nematoda class and Ascarididae family. It is a chronic parasite and, in some cases, fatal. The human disease produced by the helminths of the *Lagochilascaris* genus – lagochilascariasis (Artigas et al., 1968) – was first described in 1909, on the Trinidad and Tobago islands (Botero & Little, 1984). Following the discovery,

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other cases were reported in Latin American countries (Campos & Freire-Filha, 1989; Queiroz-Telles & Salvador, 2019). The first description in Brazil occurred 60 years later in the State of São Paulo (Campos et al., 1988). The appearance in the Brazilian Amazon, where the highest incidence has been detected, especially in the States of Acre, Pará, Roraima, Rondônia and Tocantins (Campos & Barbosa, 2016), was initially reported in 1978 (Campos et al., 1992). However, recently, a case of the disease was reported in the Brazilian mid-west (Queiroz-Telles & Salvador, 2019). The illness has since been considered an emerging zoonosis due to the rising incidence in these tropical regions. It should be noted that an emerging infectious disease “is one that appeared in a population for the first time, or that has existed previously, but is rapidly increasing in incidence or geographic range” (Ogden et al., 2017).

The life cycle of the parasite is still little known, as well as its transmission mechanism. Some studies suggest that wild carnivores are the natural hosts, while man and domestic animals, such as cats and dogs, can become infected and serve as accidental definitive hosts (Barbosa et al., 1998). The infection has greater prevalence within lower class individuals that are more vulnerable due to poor living conditions, particularly in the rural areas. The majority of those infected reside in small deforested areas, where people normally eat armadillos and guinea pigs, animals that have been speculated as being intermediate or paratenic or transport hosts of *Lagochilascaris* (Campos & Barbosa, 2016). The main signs reported in cases of *lagochilascaris* affecting *Homo sapiens sapiens* are related to the appearance of subcutaneous purulent lesions in the neck, mastoid, ear and pharynx (rhino and oro) regions (Mendes et al., 2018).

The purpose of this review is to systemize information on the disease, whose management is still scarce in medical literature. The elucidation of the pathophysiology, clinical aspects, treatment, prophylaxis and control of this morbid condition would ensure proper guidance for health professionals, in order to prepare them to treat the cases of this parasitic disease successfully.

## ETIOLOGY AND EPIDEMIOLOGY

Currently, six nematode species (*L. minor*; *L. major*; *L. turgida*; *L. sprenti*; *L. multipapillatum* and *L. buckleyi*) are known from the *Lagochilascaris* genus. *Lagochilascaris minor* is considered the etiological agent of the disease in humans (Queiroz-Telles & Salvador, 2019; Campos et al., 2017) and is the only one that has been found infecting domestic animals: dogs and cats. Other two species parasitize marsupials and two parasitize wild felids (Bento et al., 1993). The adult *L. minor* have a milky white coloring, and their bodies present lateral fins. The male worms are approximately 9 mm in length and 0.4 mm in diameter and are smaller than females which measure approximately 15 mm in length and 0.5 mm in diameter (Artigas et al., 1968). The mouth is located in the anterior region of the body along with three large and well-developed lips, separated

by intermediate lips; as well as a deep median groove (Artigas et al., 1968). The eggs have a rounded shape and measure between 50 and 90  $\mu\text{m}$  (Queiroz-Telles & Salvador, 2019). They present a thick external shell with multiple grooves and a characteristic “bottle cap” aspect. These peripheral grooves are of taxonomical value and the presence of less than 25 is indicative of the species *L. minor* (Artigas et al., 1968). The adult worms - male and female - and the eggs can be found within the lesions in vertebrate hosts.

By 2017, 128 cases were reported worldwide, 100 of which in Brazil (Barreto et al., 2018). This small number, which may be due to misdiagnosis and/or lack of notification, indicates insufficient data for lagochilascaris to be considered a relevant health problem (Artigas et al., 1968). However, this parasitic disease is seen as an emerging disease in the American continent and affects underprivileged individuals, especially those in the rural areas. Regarding distribution, human cases have been reported in neo-tropical regions, in countries such as Bolivia, Brazil, Colombia, Costa Rica, Mexico, Trinidad and Tobago, Venezuela and Suriname (Douma et al., 2016). The Amazon region is important regarding the occurrence of helminthiasis, presenting the majority of Brazilian cases, particularly the regions located between the Tocantins and Araguaia rivers (Campos & Barbosa, 2016). In the literature that described Brazilian cases, the majority (60%), were reported in the State of Pará, followed by Rondônia, and Tocantins, both in the Amazon region (Bento et al., 1993).

To clarify the biological cycle and the parasite transmission mechanism of *L. minor*, many studies have been performed involving experimental mice and domestic cat infections. Larvae hatch in the mouse gastrointestinal tract a couple of hours after infection through oral transmission of nematode eggs. After migrating through the liver and the lungs, the nodules containing the encysted larvae present an irregular distribution in the cervical, thoracic, abdominal, lumbar and axillary regions, as well as the paws. Cats are not infected after egg ingestion since helminths cannot achieve sexual maturity in these animals. Therefore, they only become infected after ingesting third stage larvae present in rodents (Campos et al., 2017). The passage of the parasite through the intermediate host contributes to the helminths' resistance and, therefore, to its development in the definitive cat host (Leão et al., 1996).

It is believed that the natural hosts (wild carnivores) harbor the adult worm in their respiratory or digestive system, where the females lay their eggs. The embryonated eggs, eliminated along with feces, contaminate the environment - water and soil (Artigas et al., 1968) - which become the source of infection for other wild animals, such as rodents (Campos et al., 2017). The ingested eggs, found in the latter, develop into their larval form and encyst themselves in muscle cellular and subcutaneous tissue known as intermediate hosts (Leão et al., 1996). When such rodents are ingested by other animals (definitive hosts), the larvae develop into adult worms, completing the natural zoological cycle (Artigas et al., 1968).

Among the hypotheses that explain the infection mechanism as well as the lesions in animals — human and non-human (domestic), the most accepted theory suggests that the infection begins from the ingestion of encysted larvae in muscles and other tissues of wild animals. Some experimental studies show that the third stage larvae, found in intermediate hosts, were able to free themselves from the cysts located in the definitive host's stomach lumen. They then migrated to structures in the neck and other adjacent areas through the esophagus, tropism that still has not been clarified (Leão et al., 1996). There are indications that auto-infection may occur, once it is possible to identify all the helminth evolutionary stages (egg, larvae and adult worms) inside the lesions (Paçô & Campos, 1998).

## PATHOGENESIS

The result of the infection process by *L. minor* is basically the appearance of a painless granulomatous reaction. In histopathological analyses, the lesions have the appearance of abscesses interconnected by fistulous trajectories, surrounded by granulated tissue, giant multi-nucleated cells and areas of fibrotic tissue. Inside these formations, it is possible to note purulent material, where parasites, in a variety of evolutionary stages, may be present (Paçô, 1994). Proteolytic enzymes in *L. minor* may facilitate their migration through the tissues of the host through the hydrolysis of extracellular matrix collagens (Bento et al, 1993). An associated bacterial infection is not uncommon. Injuries are found in places such as mastoids, palatine tonsils, eyeballs, neck, facial sinuses, middle ear, central nervous system, lungs, rhinopharynx and dental alveoli (Douma et al., 2016). However, the most afflicted areas are the neck (59.7% of the cases), mastoid (35.5%) and middle ear (29.0%) (Barbosa et al., 1998). However, the lesions can extend further than these structures, invading neighboring regions with access to the digestive tract and upper airway, affecting the rhinopharynx, oropharynx, sinuses, tonsils, dental alveoli, auditory tubes and lungs, as well as the central nervous system and the eyes (Campos & Freire-Filha, 1989). Sacral compromise has already been described. There are also reports of immunocompromised patients with the disease (Palheta-Neto et al., 2002).

## CLINICAL ASPECTS

In humans, the disease normally begins in an insidious manner, with a chronic nature along with remission and recurrence periods. The clinical presentation depends directly on the location of the lesions. Generally, there is involvement of the cervical regions with nodule fixation in deep planes that may develop into fistulas or ulcers and abscesses, as well as suppurative otitis and mastoiditis (Artigas et al., 1968). Such lesions can even expel worms measuring between 5 to 15 mm (Queiroz-Telles & Salvador, 2019).

The clinical condition may be compromised with associated weight loss. When the central nervous system is affected, convulsions, cerebellar syndrome and paralysis of cranial nerves may occur leading to death (Roig-Ocampos et al., 2010). Death by pulmonary impairment has been reported (Paçô & Campos, 1998). Another common patient complaint is the elimination of live parasites from lesion orifices, or from the external auditory tubes, mouth or nostrils (Campos & Freire-Filha, 1989).

Chronic headache and diarrhea, cachectic aspects, and stiff neck are symptoms described. The complete blood count presents slight microcytic anemia, thrombocytosis and eosinophilia. Cervicalgia does not respond to common analgesics, hardly responding to meperidine (Douma et al., 2016). Seizures, headaches, paresthesias, motor alterations and mental confusion are some phenomena observed and related to neurological impairment, along with vomiting and facial paralysis. Bilateral bronchopneumonia with fever and dyspnea, are also described (Bento et al., 1993).

## DIAGNOSIS

In clinical practice, the correct diagnosis of lagochilascariasis is of paramount importance, in order to differentiate it from other diseases to prevent the onset of severe illnesses that affect the lungs and the central nervous system (Barreto et al., 2018). The etiological diagnosis is made by visualization (macro and/or micro) of the adult worm, the larvae or the eggs in the purulent secretions originating from the lesions (Artigas et al., 1968). Adult worms and eggs can be observed in rhinopharynx or oropharynx secretions; moreover, adult helminths can also be seen leaving the oral cavity (Paçô, 1994). However, when the lesions trigger ulceration and no purulent material is produced for analysis, due to the nodules being recent or to involvement of other regions (lungs, nervous systems, etc.), the diagnosis becomes more difficult (Paçô, 1994).

When the lesions appear in the digestive tract, it is possible to find *L. minor* eggs in the patients' feces (Artigas et al., 1968). Radiological methods can be used to evaluate the extension of the inflammatory process in different organs and systems. Chest telerradiography is used to evaluate pulmonary ailment while computerized tomography is applied in cases when mastoid and central nervous system are affected (Queiroz-Telles & Salvador, 2019). Regarding cerebral lesions, the suspected diagnosis can be equally established through imaging exams (Paçô, 1994); however, the etiological diagnosis will depend on the visualization of the helminth (by biopsy, for example). The collected worms can later be viewed by scanning electron microscopy (SEM) and optical microscopy. Skin biopsy shows parasite eggs presenting thick skin and coarse surfaces measuring from 50 to 90 µm. Adult worms observed by optical microscopy present, at the rear, the ejaculatory duct, which is surrounded

by spikes. (Queiroz-Telles & Salvador, 2019). These measure 5 to 20 mm and are milky-white color (Barreto et al., 2018).

The differential diagnosis includes pyogenic adenitis, leishmaniasis, tuberculous lymphadenitis and other diseases presenting clinical conditions similar to those noted in *L. minor* infections. When larvae are observed in the lesions, the differentiation between *L. minor* and arthropod larvae that cause myiasis becomes necessary (Campos & Freire-Filha, 1989).

Complementary exams, such as complete blood workups, present little relevance in the diagnosis of helminthiasis and are not specific (Queiroz-Telles & Salvador, 2019). They can reveal leukocytosis or leukopenia, eosinophilia and even aneosinophilia (Artigas et al., 1968). Immunological exams, used to diagnose numerous parasitic infections, are still not part of the routine lagochilascaris diagnosis (Paçô, 1994). However, from a diagnostic point of view, the work of Prudente et al. in 2009 revealed that the gross *L. minor* extract can be considered a good antigen and that the detection of IgM and IgA in ELISA trials can indicate recent infection while the detection of IgM and IgG may suggest a chronic process.

## TREATMENT

Helminthiasis treatment becomes more difficult due to the heterogeneous aspect of the parasite's cycle and by its wide and variable clinical spectrum (Semerene et al, 2004). Since the first reports of lagochilascaris, many therapeutic regimens have been used: cambendazole, thiabendazole, levamisole, diethylcarbamazine, mebendazole, albendazole, praziquantel and ivermectin; and many others have been tested. However, these treatments proved unsuccessful due to frequent recurrences after months and even years of the apparent clinical cure, a situation that highlights the need for prolonged follow-up periods after treatment (Smith et al., 1983). The success of the therapeutic approach is only possible with the use of a drug or a pharmacological association that affects all of the evolutionary phases of the helminth (adult worm, larvae and egg), preventing the continuity of the cycle (Semerene et al., 2004). The best clinical results in human beings have been attained by using 20 to 30 mg/kg/day of cambendazole, especially when associated with levamisole (80-150 mg/day for 3 days) during the first dose (Campos & Freire-Filha, 1989). These drugs act directly upon the parasite, which constitutes a pharmacological advantage (Artigas et al., 1968). It should be noted that, due to recurrences, the adoption of a management regime, utilizing cambendazole (new series every 6 months), or levamisole (every 6 months), is recommended. Diethylcarbamazine can also be used from six months to a year, during long-term treatment (Artigas et al., 1968).

Another medication that is being used, in an experimental manner, is ivermectin (Sudré et al., 2012). There are reports of success with the use of the drug in therapies where cambendazole and levamisole were suspended due to intolerance (Vargas-Ocampo & Alvarado-Aleman, 1997).

The removal of nodules is indicated to reduce the healing time. Nevertheless, this procedure can be used as an auxiliary resource to pharmacological therapy (Queiroz-Telles & Salvador, 2019).

## PROPHYLAXIS AND CONTROL

Considering the paucity of relative information on epidemiology and the mechanisms of infection of *L. minor*, the determination of effective prophylactic measures has become a great challenge. Education regarding the danger of eating rodent meat, which is an intermediate host, is needed in public health and prevention strategies. Rats, guinea pigs, and agoutis are often used as food in these regions being, therefore associated with the incidence of human lagochilascariasis. In the midst of the little data that exists, some recommendations include: (1) avoiding the consumption of uncooked or rare meat, rodents in particular, and (2) drinking only filtered water (Volcán & Medrano, 1990).

## FINAL CONSIDERATIONS

Although *Lagochilascaris* was first described at the beginning of the 20th century, it is still little addressed in present literature. This lack of data can be understood due to the low incidence of the disease as well as the under-reporting of cases. The shortage of information prevents the further development of knowledge about the peculiarities of the disease, including the etiological agent's biological cycle and clinical manifestations.

Considering what is known about the illness, it is important to prepare health professionals so they are aware of this possibility when seeing individuals with granulomatous reactions in the cervical, mastoid and middle ear areas, as well as enabling them regarding diagnostic investigation and in the choice of the appropriate therapeutic regimen (once lagochilascaris is confirmed). Furthermore, instructing patients regarding prevention and health measures should be further discussed, especially the population living in rural areas that are considered at risk of contamination by this helminth.

## CONFLICT OF INTEREST.

There is no conflict of interest.

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