

HUMAN LAGOCHILASCARIASIS TREATED SUCESSFULLY WITH IVERMECTIN: A CASE REPORT

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SUMMARY

A 15 year old male patient was diagnosed as being infected with the nematode *Lagochilascaris minor*, presenting as abscesses over the left mastoid region, and invading the temporal bone, mastoid sinuses and possibly the CNS. Surgical drainage and administration of cambendazole and levamisole led to improvement, followed by an early relapse and poor tolerance to the antihelminthic drugs. Repeated doses of ivermectin (an animal preparation) were used for the first time to treat this condition and resulted in complete remission of signs of infection, maintained for 15 months after the end of drug therapy, indicating cure of the disease.

KEY WORDS: *Lagochilascaris minor*; Ivermectin; Human infection; Treatment.

INTRODUCTION

Lagochilascariasis is a rare and poorly known zoonosis caused by *Lagochilascaris* sp., an ascarid nematode usually shorter than 26 mm. Five species have been described: *L. major* Leiper, 1910, and *L. buckleyi* Sprent, 1971 in felines, *L. turgida* (Stossih, 1902) Travassos, 1924, *L. sprenti*. BOWMAN et al., 1983 in marsupials, and *L. minor* Leiper, 1909 in dogs, cats and human cases².

L. minor is considered an abnormal parasite of humans. The infection appears usually as tumoral lesions, such as nodules, pseudocysts or abscesses in the cervical region. Invasion of paranasal sinuses, dental alveoli, oro and rhinopharynx, ear, eye and CNS structures have been described^{2,5,8,9}, as well as pulmonary lesions^{11,13}. All stages of the parasite are found in tissues: eggs, larvae and adult worms; they are eliminated spontaneously through fistulae draining to the skin or pharyngeal mucosa. Local reproduction and auto-infection is thought to occur and maintain the infection for long periods^{4,9}.

Geographic distribution of *L. minor* is reported to be exclusively neotropical. FRAIHA et al., 1989⁹ have published an extensive review of the disease, gathering 62 reported cases. Brazil accounts for 74% of the cases, most occurring near the rivers Tocantins and Araguaia, in the Center-West and North of the country; cases were also reported from Colombia, Venezuela Trinidad, Tobago, Suriname, Costa Rica and Mexico. Life cycle and mode of infection remain unknown, but contact with larvae eliminated by wild felines may be an important way of transmission. FRAIHA et al.⁹ and others^{8,15} favor the hypothesis that after ingestion, the larvae would reach the lungs, similar to *Ascaris lumbricoides*, but instead of entering the digestive tract, they would penetrate the pharyngeal mucosa and develop in the nearby structures.

Surgical drainage of the abscesses are mandatory, and adjunctive drug treatment has led to erratic and often unsatisfactory response. Prognosis is poor if essential structures are compromised; five cases of death have been

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reported⁹. Several drugs have been used, with variable efficacy: levamisole, thiabendazole, cambendazole, mebendazole, albendazole, praziquantel, and diethylcarbamazine⁹. Concomitant use of cambendazole and levamisole have been recommended as the most effective regimen⁸. We report a case with extensive cervical lesions, that responded well to therapy, and relapsed later, achieving total remission after repeated doses of ivermectin.

CASE REPORT

In December of 1990, a 15 year - old male patient from Cruzeiro do Sul, Acre state, in the North of Brazil, was referred to our Ear Nose and Throat clinic in São Paulo, because of chronic fetid ear secretion and recent bulging of the retroauricular region. On the examination, he was in good clinical condition, had unilateral purulent and fetid otorrhoea and an inflammatory tumor over the left mastoid region, approximately 9 by 6 cm, with two draining fistulae.

Computerized tomography showed invasion of the middle and posterior cranial fossae, osteomyelitis of the temporal bone and extensive abscess of the cervical region, but no clinical signs of neurological involvement. Surgical drainage was performed, when small whitish organisms, measuring approximately 10 mm were seen in the pus, and identified as *Lagochilascaris minor*.

The patient was treated with antibiotics and anti-helminthic agents. Cambendazole (four cycles of 30 mg/Kg daily for five days, repeated after a 10-day interval) and levamisole (150 mg daily for 10 days, then 150 mg once a week for 3 months) were given. Surgical drainage was repeated when purulent secretion was found clinically to accumulate. Good immediate clinical response was observed, with closure of the fistulae and complete regression of the tumor at the end of the proposed drug regimen.

One month after the end of drug therapy, he was admitted again with pain over the left mastoid and a 15 cm abscess at the same region, draining pus by several fistulae; *L. minor* larvae were identified again in the pus. Cambendazole and levamisole were resumed, but the patient developed severe gastrointestinal symptoms and hair loss, after a few days; liver enzymes were also elevated. All signs disappeared following drug discontinu-

ation. Abscess drainage was performed many times. Ivermectin was then administered, with the consent of the patient in two cycles of four doses of 0.2 mg/Kg at weekly intervals, followed by a month without therapy. As no human preparation was available at the time, we used an animal preparation — Oramec, MSDAgVet — indicated for oral use in sheep. Clinical tolerance, blood and urine chemistry and counts were closely followed during therapy. Symptomatic relief was noted after the second dose, and secretion output was reduced; no other surgical drainage was necessary. After the second cycle of ivermectin, clinical signs of infection had subsided completely; monthly doses of ivermectin were given thereafter for six months. No clinical or laboratory side effects were observed. Fifteen months after therapy discontinuation, there was no evidence of recurrence of the disease; clinical examination of the cervical region showed only moderate fibrosis below the left mastoid.

DISCUSSION

Lagochilascariasis is a rare parasitosis occurring in South America, and probably underdiagnosed, since most patients come from rural poor areas, where medical care is scarce. Clinical features are still intriguing, and deserve further study, considering the seriousness and even fatal outcome of many cases. Both drug and surgical therapy seem to be important to control cervical lesions. Pyogenic abscess is the most common initial diagnosis, demanding surgery; detection of worms in pus or histological examination is often the clue to the definition of the parasitic disease. Drug treatment has been not been reported to be used alone, but its importance as an adjunct to surgery is unquestionable. The chronic and recurrent nature of the infection and difficulties to define parasitological cure in closed lesions, however, have impaired the establishment of optimal drug therapy.

The present case was treated as recommended by FRAIHA et al.⁸, with a combination of cambendazole, an imidazole derivative active against *Strongyloides stercoralis*, and levamisole. Good efficacy was observed at the beginning, but soon afterwards the disease recurred, and a second trial of drug therapy had to be discontinued because of poor tolerance. Since other options were not associated with good efficacy, and the patient had evidence of imminent invasion of CNS, an alternative drug was considered.

Ivermectin is a macrocyclic lactone derived from a group of naturally occurring substances called avermectins, widely used in agriculture and animal diseases⁷. It has a wide spectrum of action against round worms and arthropods¹⁶, and has been used successfully in humans for the treatment of filariasis and intestinal helminthiasis.^{1,10,12} Despite no previous published experience in lagochilascariasis, it was chosen to be used here because of its long half-life and wide distribution in the body⁶, allied to a previous report of in vitro activity against larvae of *L. minor*, at small concentrations⁴. A regimen of repeated doses was prescribed based on the possibility of autoinfection and the chronic course of the disease.

The results obtained in the present case were indicative that ivermectin is useful for the treatment of lagochilascariasis; however, it is impossible to ascertain the most appropriate therapy schedule. The use of several doses seemed to be beneficial. Another case reported to us received one single dose of 3 mg of ivermectin and had only temporary closure of the fistulae (SILVEIRA, CAN — not published). Further clinical and experimental observations regarding the efficacy of this drug against this parasite appear to be justified.

RESUMO

LAGOCHILASCARIÁSE HUMANA TRATADA COM SUCESSO COM IVERMECTINA: UM RELATO DE CASO.

Um jovem de 15 anos, masculino recebeu o diagnóstico de infecção pelo nematódeo *Lagochilascaris minor*, manifesta na forma de abscessos sobre a região mastoidea e invadindo o osso temporal, células mastoideas e, possivelmente, o SNC. Foi feita drenagem cirúrgica e administrado cambendazole e levamisole, seguidos de melhora e posterior recaída; ocorreu também intolerância aos anti-helmínticos. Doses repetidas de uma preparação animal de ivermectina foram utilizadas pela primeira nesta doença, levando a remissão completa dos sinais de infecção, mantida após 15 meses do final do tratamento e indicando cura do processo.

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