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Angiostrongylus costaricensis: FIRST RECORD OF ITS OCCURRENCE IN THE STATE OF ESPIRITO SANTO, BRAZIL, AND A REVIEW OF ITS GEOGRAPHIC DISTRIBUTION.

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SUMMARY

Abdominal angiostrongyliasis is a parasitic disease caused by *Angiostrongylus costaricensis*, a metastrongylid nematode with wide geographic distribution, occurring from the United States to Argentina. In Brazil, the disease has been reported from the States of Rio Grande do Sul, Santa Catarina, Paraná, São Paulo, Federal District of Brasília and Minas Gerais. We report here a case of abdominal angiostrongyliasis in a 9-year-old girl, from Itatiba, State of Espirito Santo, Brazil, submitted to exploratory laparotomy for acute abdomen. Extensive inflammatory lesions of terminal ileum and cecum, with perforations of the first, were present, and ileocecal resection was performed. The pathological picture was characterized by transmural inflammatory granulomatous reaction, extensive eosinophilic infiltration, eosinophilic vasculitis and the presence of worms within a mesenteric artery branch, with histological features of metastrongylid nematodes. This case report contributes to a better knowledge of the geographic distribution of this parasite in Brazil, suggesting that abdominal angiostrongyliasis may represent a disease of medical importance, more than a rarity of academic interest.

KEYWORDS: *Angiostrongylus costaricensis*, Abdominal angiostrongyliasis; Geographic distribution.

INTRODUCTION

Abdominal angiostrongyliasis is a nematode parasitic disease caused by *Angiostrongylus costaricensis*[43]. This metastrongylid nematode was described by MORERA and CESPEDES (1971)[44], in Costa Rica, where the classical clinical-pathological syndrome is known to occur since 1952[9]. The clinical picture is characterized by abdominal pain in the right iliac fossa, anorexia, vomiting, diarrhoea and constipation [30,31,47]. On palpation of the abdomen there is pain, and at least half of the cases a tumour-like mass can be observed on the right lower quadrant[31]. Laboratory findings include leukocytosis with eosinophilia[16,31].

In the definitive host, *Angiostrongylus costaricensis* is found in the branches of the mesentery artery irrigating the terminal ileum, appendix and cecum[37]. Natural definitive hosts include various rodents (*Sigmodon hispidus*[36,52,57,59] *Rattus rattus*[36,57], *Zygodontomys microtinus*[57], *Liomys adspersus*[57], *Oryzomys fulvescens*[57], *O. caliginosus*[34], *O.nigripes*[15], *O. ratticeps*[15], *Proechimys sp*[51]) and other mammals (*Saguinus mystax*[55], *Nasua narica bullata*[35]). Eggs deposited in the mesenteric arteries are carried with the blood flow to the capillaries in the intestinal wall, develop to larvae which, after hatching, migrate through the tissues, reach the intestinal lumen and are

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evacuated in the faeces[37]. The larvae are ingested by intermediate hosts, veronicellid slugs (*Vaginulus plebeius*[13,42,53], *Phyllocaulis variegatus*[21], *Phyllocaulis soleiformis*[19]) and other mollusks (*Limax maximus*, *L. flavus* and *Bradybaena similaris*)[20], where they become infective to the mammal host. The definitive host acquires infection by ingestion of the slugs or food contaminated with infective larvae shed by their mucus[37].

Man is an accidental host and probably acquires the infection by ingestion of fruits and vegetables contamined with infective larvae. The majority of the lesions occur in the terminal ileum, appendix and cecum[9,31]. The pathological picture is characterized by exuberant tissue eosinophilia, oedema of all layers of the intestinal wall, granulomas with eggs, some with developing larvae, consisting of macrophages, a few lymphocytes and abundant eosinophils[9,18]. The vessels containing adult parasites may show eosinophilic infiltration; sometimes they are thrombosed and may contain a dead worm. The presence of the worm is not required for the diagnosis, and the detection of massive eosinophilic infiltration in all layers of intestinal wall, granulomatous reaction and eosinophilic vasculitis affecting arteries, veins, lymphatic vessels and capillaries, establish the diagnosis of probable abdominal angiostrongyliasis[18]. In human infections, larvae have not been detected in faeces, since their elimination is prevented by the inflammatory reaction in the intestinal wall. In rare cases, *Angiostrongylus costaricensis* has been found in the liver [46] and in spermatic artery [49]. The diagnosis relies mainly on histopathological examination of biopsies and surgical specimens. Serological tests are used in Costa Rica[38].

We report here a case of abdominal angiostrongyliasis from the State of Espirito Santo, Brazil, contributing, this way, to a better knowledge of the geographic distribution of *Angiostrongylus costaricensis* in Brazil, and reinforcing its importance in the pathology of this country.

CASE REPORT

A 9-year-old girl was submitted to exploratory laparotomy for acute abdomen, in Manhuaçu, MG, in March, 1993. She lived in the rural area of Itatiba, Espirito Santo, and went to Manhuaçu only to receive medical care. The clinical picture was reported to be so dramatic that no pre-operatory laboratory studies could have been performed. Surgical findings included extensive inflammatory lesions in terminal ileum, with perforations, and diffuse mesenteric lymphadenitis. Enteric resection was performed and the surgical specimen sent to the Pathology Service of Hospital Felicio Rocho in Belo Horizonte, with suspected diagnosis of ileitis or tuberculosis. The surgical specimen consisted of 11cm of terminal ileum, appendix and 15 cm of cecum and ascending colon. Marked thickening of colonic wall close to ileocecal valve was observed. The terminal ileum showed moderate dilation of the lumen, thickening of the wall and areas of perforation (Fig. 1). Mesenteric lymphadenectasis was also observed.

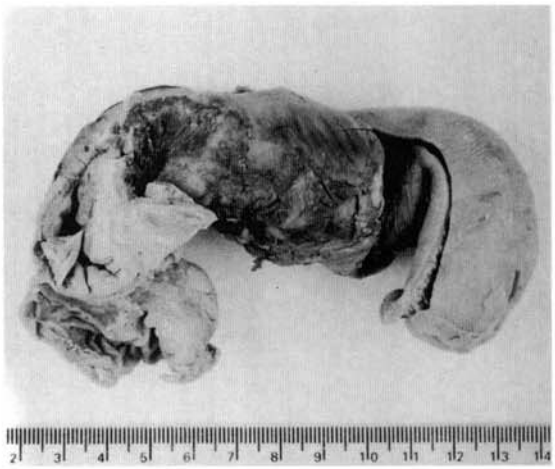

Fig. 1 - Macroscopical aspect of the specimen. Markedly thickening of the intestinal wall, close to ileo cecal valve (left side) is observed. Ileal segment showing extensive hemorrhage, dilation of the lumen and thickening of the wall (original magnification 75 x)

The histological study of the thickened colonic wall showed massive granulomatous inflammatory reaction, with intense eosinophilia (Fig. 2). Vascular changes were present and consisted of thrombosis and eosinophilic infiltration of vascular wall (Fig. 3). Worms with histological characteristics of metastrongylid nematodes were found in only one arterial vessel (Fig. 4), although several sections had been undertaken. No larvae or eggs could be identified. The terminal ileum presented severe inflammatory changes, with necrosis and perforations; in this segment, extensive eosinophilic infiltrate and granulomas were also observed. Appendix and lymph nodes showed reactive

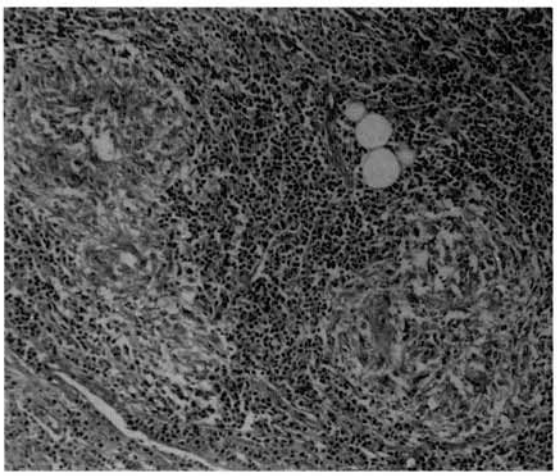

Fig. 2 - Granulomatous reaction and extensive inflammatory infiltrate, which is composed mainly of eosinophils (original magnification 75 x)

Fig. 3 - Vascular changes in abdominal angiostrongyliasis. Arterial vessel with lumen occupied by a thrombus. Partial destruction and inflammatory infiltration of vascular wall (original magnification 75 x)

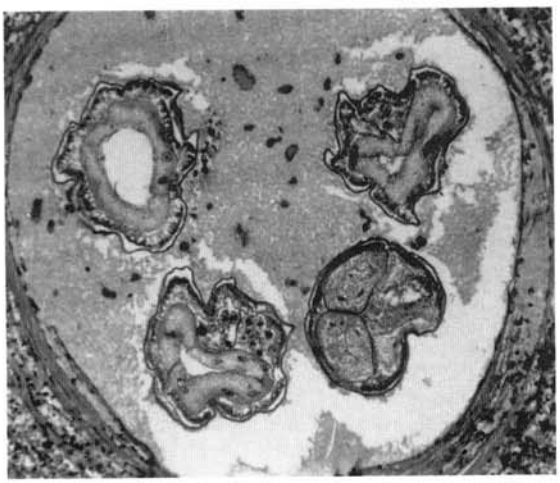

Fig. 4 - Arterial vessel with four sections of worms, with histological features of metastrongylid nematodes, some showing degenerative changes; intestine showing 2 to 8 nuclei in these sections; intestinal cell boundaries are not discernible. Muscular layer partially detached from cuticle; muscle cells not well defined. Lateral cords are inconspicuous. Structure of gonad also seen (original magnification 75 x)

changes, characterized by hyperplastic lymphoid follicles, and sparse neutrophilic and eosinophilic infiltration.

After two weeks of hospitalization, she was discharged, aparently cured. It was not possible to obtain any follow-up information.

DISCUSSION

The diagnosis of abdominal angiostrongyliasis in this case was made on the basis of the histopathological lesions, characterized by transmural inflammatory granulomatous reaction, dense eosinophilic infiltration, eosinophilic vasculitis and the presence of nematode parasites within mesenteric artery branch. The localization of the parasite and consequent histopathological lesions correspond to those described in the literature[9,18]. Our case presented with the appearance of a pseudoneoplastic lesion[18]. The intestinal wall close to ileocecal valve was enormously thickened and constricted the lumen. Our patient was 9 years old, within the age group most commonly affected by the disease in Central America[9,30,31,47]. In Brazil, however, both adults and children are affected [1,4,16,23].

The great majority of cases reported in literature are from Costa Rica [9,30,31,47], where prevalence has been estimated to 12 cases per 100,000 habitants per year[38]. In this country, the most important intermediate hosts are veronicellid slugs, an agricultural pest in Central America, which are found infected at high rates[11]. The cotton rat (*Sigmodon hispidus*) is the most important definitive host in Central America [38].

Human cases of the disease has also been reported from the United States[22,27,54], México[60], Honduras[53,62], El Salvador[52], Nicaragua[14], Panama[50], Martinica[24], Guadaloupe[25], and Argentina[12]. Human disease is known to occur in Venezuela, Colombia and Guatemala[40]. Occurrence of the parasite has been reported from Peru[55], where definitive hosts were found infected. Infection of rodents and other mammals has also been reported from United States[59], El Salvador[52], Panama[57], Colombia[34] and Venezuela[51]. In Ecuador, *A. costaricensis* has been recovered from slugs of the genus *Vaginulus*[45]. *A. costaricensis* has also been detected in intermediate hosts from Honduras[26,41,53] and Nicaragua[13]. A case of abdominal angiostrongyliasis in Africa has been reported [6].

In Brazil, to the present, 42 cases have been reported, including the case reported here: 27 cases from the State of Rio Grande do Sul [1,2,16], four cases from the State of Santa Catarina [4,32], four cases from the State of Paraná[4,5], three cases from the State of São Paulo[8,23,61], two cases from the Federal District of Brasilia [7,33], one case from the State of Minas Gerais[48] and this one from the State of Espirito Santo. In the State of Rio Grande do Sul, most cases have been found in a survey made in Pathology Services[16]. In Southern Brazil, naturally infected slugs[19,20,21] and rodents[15] have been detected. Recently, it has been demonstrated that specimens of *Sarasinula marginata*, collected in kitchen and house gardens in Belo Horizonte, MG, are susceptible to experimental infection with *Angiostrongylus costaricensis*[28]. No information about intermediate and definitive hosts from other areas of Brazil is yet available. Species of *Biomphalaria*[29,56,58] and *Veronicella occidentalis*[34] are also known to be susceptible intermediate hosts, at least under laboratory conditions.

Although its wide geographic distribution, the parasite has not yet been detected in other Brazilian states.

Abdominal angiostrongyliasis may probably represent an under diagnosed disease by virtue of (1) absence of serological diagnosis; (2) absence of larvae in human faeces; (3) many surgical specimens, specially those diagnosed clinically as "appendicitis" are not sent to pathological study; and, finally, (4) the unawareness of the disease among medical personnel[17]. Other possible source of confusion may be the presence of *Enterobius vermicularis* in appendix specimens[10]. Confusion should not be made with *Schistosoma mansoni*, another intravascular helminth, with distinct morphology, which is found in venous vessels and produces eosinophilic granulomas containing characteristic eggs [9,17]. Differential diagnosis in surgical specimens should also include anisakiasis[3].

It has been postulated that current knowledge of the geographical distribution of abdominal angiostrongyliasis represents only the tip of the iceberg, since the differences between recorded prevalence in Costa Rica and that of other Central and South America countries are hard to explain in the light of special environmental conditions in that country[38]. The possibility that this zoonosis occurs in wild and peridomiciliary cycles may explain some of the differences of prevalence of human disease in various areas. Available data of definitive and intermediate natural hosts suggests that the biological cycle of *A. costaricensis* involves various mammals and mollusks[39], some of them occuring in large numbers in cultivated agricultural and peridomiciliary areas, providing many opportunities for humans to become infected. On the other hand, natural infection of wild mammals has also been reported[35,55]. The case reported here suggests that the distribution of *Angiostrongylus costaricensis* in Brazil is wider than already known, reinforcing the fact that special attention should be paid to this parasite in Brazil.

RESUMO

Angiostrongylus costaricensis: primeiro relato de sua ocorrência no estado do Espírito Santo, Brasil, e revisão de sua distribuição geográfica.

Angiostrongylus costaricensis, um parasita nematódeo, com ampla distribuição geográfica, é o agente causal da angiostrongilíase abdominal. No Brasil, a doença tem sido descrita nos Estados do Rio Grande do Sul, Santa Catarina, Paraná, São Paulo e Minas Gerais e no Distrito Federal. Relatamos a ocorrência de um caso de angiostrongilíase abdominal no estado do Espírito Santo, Brasil. Paciente de 9 anos de idade, sexo feminino, foi submetida a laparotomia exploradora por abdômen agudo. Foram observadas lesões inflamatórias extensas de íleo terminal e ceco, com perfuração do íleo, tendo sido realizada ressecção do segmento acometido. O estudo anatomopatológico mostrou inflamação granulomatosa transmural, infiltrado intenso de eosinófilos, vasculite eosinofílica e a presença de parasitas, com características histológicas de nematódeos metastrongilídeos no interior de ramo da artéria mesentérica. Nosso relato contribui para uma melhor definição da distribuição geográfica desse parasito no Brasil, evidenciando assim a importância médica dessa parasitose, anteriormente considerada como uma raridade de interesse apenas acadêmico.

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