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CASE REPORT

Pulmonary paragonimiasis: report of a case in resection specimen and review of distribution in Colombia

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Abstract

Paragonimiasis constitutes a parasitic disease caused by trematodes of worldwide distribution. More than 30 species of these parasites that infect animals and humans; *Paragonimus spp*. predominate in America, Africa, and Southeast Asia.

A 44-year-old female patient from Valle del Guamuez is presented. Since 1 year ago she presented cough without expectoration, in the last 3 months associated with hemoptotic traces; and since 2 weeks ago with frank hemoptysis and pain in left hemithorax without other associated symptomatology. Chest CT scan showed an area of cavitated consolidation located in the upper segment of the left lower lobe. She was taken to segmental lobectomy. Histological sections showed lung with a pattern of lymphohistiocytic inflammation with formation of abundant granulomas, some of them with necrosis. Inside the granulomas, thick-walled parasitic eggs were identified, birefringent under polarized light, measuring between 50 and 80 um, which allowed the diagnosis of pulmonary Paragonimiasis.

This work proposes the publication of a case of paragonimiasis in humans diagnosed for the first time in a surgical resection specimen, a review of the literature and of the cases previously described in Colombia, its geographical distribution, and associated variables.

Keywords: Paragonimus; Paragonimiasis; eosinophilia; pulmonary stave; Colombia

Paragonimiasis pulmonar: reporte de un caso en pieza de resección y revisión de su distribución en Colombia

Resumen

La paragonimiasis es una enfermedad parasitaria causada por trematodos de distribución mundial. Existen más de 30 especies de estos parásitos que infectan a animales y humanos; Paragonimus spp. predomina en América, África y el Sudeste Asiático.

Se presenta el caso de una paciente de 44 años del Valle del Guamuez. Desde hacía un año presentaba tos sin expectoración, asociada en los últimos 3 meses a trazas hemoptóticas; y desde hacía 2 semanas, hemoptisis franca y dolor en el hemitórax izquierdo sin otra sintomatología asociada. La tomografía computarizada de tórax mostró un área de consolidación cavitada ubicada en el segmento superior del lóbulo inferior izquierdo. Fue sometida a una lobectomía segmentaria. Los cortes histológicos mostraron un pulmón con un patrón de inflamación linfohistiocítica con formación de abundantes granulomas, algunos de ellos con necrosis. Dentro de los granulomas se identificaron huevos parasitarios de pared gruesa, birrefringentes bajo luz polarizada, con un tamaño de entre 50 y 80 µm, lo que permitió el diagnóstico de paragonimiasis pulmonar.

Este trabajo propone la publicación de un caso de paragonimiasis en humanos, diagnosticado por primera vez en una pieza de resección quirúrgica, la revisión de la literatura y de los casos previamente descritos en Colombia, su distribución geográfica y variables asociadas.

Palabras clave: Paragonimus; paragonimiasis; eosinofilia; bastón pulmonar; Colombia

Introduction

Paragonimiasis is a parasitic disease caused by trematodes distributed worldwide. There are more than 30 species of these parasites that infect animals and humans. In the Americas, Africa, and Southeast Asia, Paragonimus spp. predominate; in Southeast Asia and Japan, Paragonimus westermani; and in North America, Paragonimus kellicotti[1]; Paragonimus mexicano is the most common in our country[2].

These parasites have a three-host life cycle. In the snail, the first intermediate host, the miracidia go through several developmental stages: sporocysts, redia, and numerous cercariae that emerge from the snail; the latter invade the second

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intermediate host, a crustacean such as the crab. In the crab, cercariae encyst and develop into metacercariae, the infective form of the mammalian host, which occurs in humans when undercooked or raw crustaceans are ingested. Metacercariae encyst in the human duodenum, penetrate the intestinal wall into the peritoneal cavity, from where they migrate through the diaphragm and into the lungs, where they encapsulate to develop into adults and secrete unembryonated eggs in the sputum and/or feces after being swallowed[1].

The clinical manifestations of infection in humans are determined by the Paragonimus species, parasite count, ploidy, and host immune status[3]. Pulmonary manifestations include cough, hemoptysis, pleuritic pain, dyspnea, and weight loss[4]; however, skin, pleural, hepatic, and cerebral involvement, among others, have also been described[2], all of which present symptoms that require consideration of tuberculosis, bacterial pneumonia, lung cancer, and even SARS-CoV-2 infection[5]. Mortality is generally low, although with a high morbidity burden[2], with an almost complete response to the oral administration of praziquantel[6].

Chest CT imaging findings in patients with pulmonary paragonimiasis include nodules, ground-glass opacities, cystic lesions with surrounding halos, tracheobronchial fistulas, fibrosis, pleural effusions, pleural thickening, calcifications, and lymphadenopathy[7].

Paraclinical examinations can identify elevated peripheral blood eosinophils, positive serum antibodies against Paragonimus as determined by intradermal test for paragonimus-specific antigens (IDTPA) and enzyme-linked immunosorbent assay (ELISA), the presence of eggs and/or parasites in bronchoalveolar lavage fluid, stool, or sputum, and, more recently, transbronchial biopsies for histopathological examination[8].

In the Cauca Valley and Colombia in general, Paragonimus has been reported since 1968, with studies of the parasite showing up to 10% prevalence in humans and up to 90% in crabs from an indigenous area in the department of Antioquia, where the main studies in the country have been conducted[9].

This article proposes the publication of a case of paragonimiasis in humans diagnosed for the first time in a surgical resection specimen; a review of the literature and previously described cases in Colombia; their geographic distribution; and associated variables.

Description of the case

We present the case of a 44-year-old female patient from Valle del Guamuez (Putumayo, Colombia), who consumes raw river shrimp during her daily tasks as a housewife. Since 1 year ago she presents cough without expectoration, in the last 3 months associated with hemoptotic traces; and since 2 weeks ago with frank hemoptysis and pain in the left hemithorax without other associated symptoms. Physical examination revealed normal vital signs and no alterations on cardiopulmonary auscultation. Chest CT scan showed an area of cavitated consolidation located in the upper segment of the left lower lobe. Blood paraclinical analysis revealed a hemogram with leukocytes 8260 cells/mm3, neutrophils 4040 cells/mm3, lymphocytes 2530 cells/mm3, eosinophils 1030 cells/mm3, basophils 50 cells/mm3, hemoglobin 13.6 g/dL, and platelets 279,000 cells/mm3. C-ANCAS (cytoplasmic pattern of anti-neutrophil cytoplasm antibodies), P-ANCAS (perinuclear pattern of anti-neutrophil cytoplasm antibodies), ENAS (extractable nuclear antigens), anti-DNA, smear microscopy and PCR for Mycobacterim tuberculosis in induced sputum were negative. ANAS were positive (1:160) with a fine granular pattern. Fibrobronchoscopy revealed evidence of acute bronchitis and clots in the left lower lobe, mild tracheobronchomalacia, and pharyngeal wall collapse, with negative microbiological studies. The cytological report of the bronchoalveolar lavage showed eosinophils at 36%.

Considering the clinical evolution and without confirmation of the etiology, the patient was evaluated by thoracic surgery, who indicated the performance of segmental lobectomy by thoracoscopy. In the pathology laboratory of the Hospital Universitario del Valle, a pulmonary wedge with usual external characteristics was processed. When cut, an abscessed cystic area of 1 cm in diameter was recognized, inside it there were two firm cylindrical structures; the perilesional parenchyma was hemorrhagic, with a hemorrhagic appearance.

The perilesional parenchyma was hemorrhagic, with hemorrhagic foci. Histological sections showed a lung with lymphohistiocytic inflammation pattern with formation of abundant granulomas, some of them with necrosis. There was scarce perilesional eosinophilia and fibrosis. Inside the granulomas there are thick-walled eggs, birefringent under polarized light, measuring between 50 and 80 um (Figure 1).

The patient evolved satisfactorily in the postoperative period and was evaluated by infectology who indicated medical treatment with albenzadol at a dose of 30 mg/kg/day for 21 days, triclabendazole was not formulated, which is the drug of choice due to its unavailability for treatment. Three months after treatment, the patient presented with improved symptoms, without confirmation of parasitological cure.

Discussion

In Colombia, the first study on paragonimiasis in humans was published at the beginning of this millennium, and no new studies have been developed to evaluate the prevalence of this disease[10]. Since 1981, 28 cases have been reported (Table 1), all diagnosed using radiological studies, intradermal tests, and/or egg characterization in sputum or fecal matter; none were diagnosed in resection specimen,s as in the case presented.

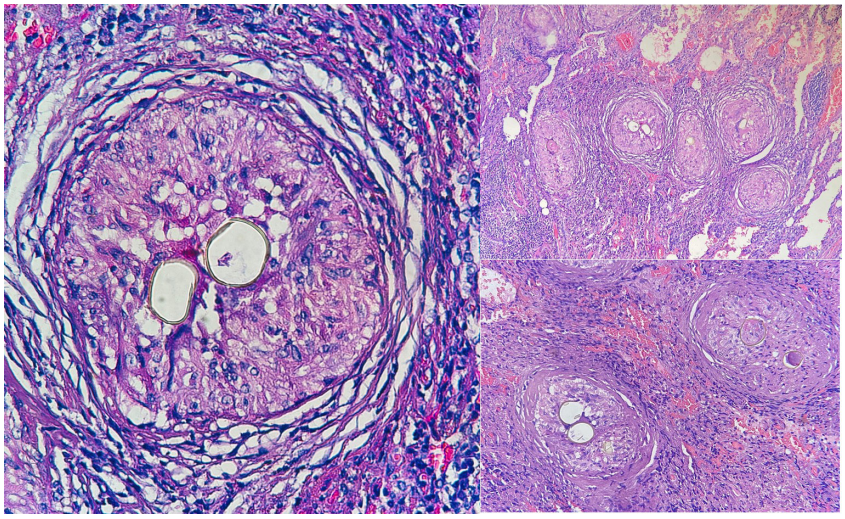

Figure 1. Hematoxylin-eosin (4X and 10X). Lung parenchyma with lymphohistiocytic inflammation pattern and formation of abundant granulomas, some of them with necrosis. In its interior there are multiple thick-walled eggs, birefringent to vision under polarized light, measuring between 50 and 80 um. At the periphery of the lesion there are abundant eosinophils.

When reviewing the distribution by sex, it is possible to identify that 60% of the cases occurred in men, 90% in the indigenous population, who also presented a comparatively younger age at the time of diagnosis than those cases in the non-indigenous population. In 100% of the cases, there was pulmonary involvement with eosinophilia as the main laboratory finding. In addition, in all patients in whom an epidemiological link was described, the ingestion of raw crabs was noted. The data described are consistent with the sociodemographic characteristics associated with this pathology[10].

When evaluating the geographical distribution, most cases were found along the Pacific coast, with only two cases in the central-eastern region of the country. This distribution is usual, as paragonimiasis is particularly frequent in tropical and subtropical regions in populations close to streams and creeks; and in populations with dietary habits of eating raw crustaceans[10]. These geographic areas with the highest number of reported cases of paragonimiasis in humans coincide with those with the highest levels of paragonimiasis in humans.

This is similar to the situation in those with the highest levels of multidimensional poverty in Colombia[10], which could be related to exposure to the parasite facilitated by housing conditions and access to public utilities.

Paragonimiasis in humans is a parasitic process that, although widely described, clinically constitutes a diagnostic challenge, as it simulates other infectious conditions, such as tuberculosis, and non-infectious conditions, such as allergic or chronic inflammatory pulmonary diseases. Currently, imaging, serological, and histological diagnostic methods are available with acceptable diagnostic performance when used in combination. In our case, the diagnostic images were inconclusive, and bronchoalveolar lavage showed eosinophilia; however, no parasite eggs were identified; therefore, direct identification in post-lobectomy tissue represents a very rare scenario, not previously reported in our country.

It is probable that the prevalence of this disease is much higher than that reported, and that it is not only distributed in areas of the indigenous population but also near large cities;

Table 1. compilation of cases of paragonimiasis in humans diagnosed in Colombia

Study	Year	Number of cases	Year	Gender	Origin	Affected organ	Population	Nexus	Laboraties	Authors
1[11]	1981	1	33	Male	Target	Lung and Liver	No date	No date	Eosinophilia	Buitrado et al
2(10)	1993-1998	24	13.8 (prom)	Male (13) Female (11)	Antioquia and chocó	Lung	Indigenous	Ingestion of crabs raw	Eosinophilia	Velez et al
3(12)	2012	1	18	Male	Antioquia	Lung	No date	Ingestion of crabs raw	Eosinophilia	Arango et al
4(13)	2013	1	40	Male	Chocó	Lung	Indigenous	Ingestion of crabs raw	Eosinophilia	Buitrago et al
5(14)	2021	1	32	Male	Bogotá D.C.	Lung, Liver and brain.	No date	No date	Eosinophilia	Donato et al

the presence of crustaceans infected with Paragonimus metacercariae, living in streams and creeks of the Antioquian peri-domicile has been previously described (9); therefore, new studies are needed for its characterization in other areas of the country.

Ethical considerations

This work was developed under the guidelines set forth in Resolution 008430 of 1993 of the Colombian Ministry of Health.

Protection of persons. Not applicable

Protection of vulnerable populations. Not applicable

Confidentiality. No names, initials or hospital medical record numbers (or any other type of irrelevant data for the research that could identify the patient) were used in either the text or the photographs, and confidentiality was guaranteed.

Privacy. Informed consent was obtained from the patient for the publication of the case and the images.

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Authors´ contribution. JSC, AFVC: Study conception and design. JSC, AFVC: Data acquisition. AC, MCM: Data analysis and interpretation. JSC, AFVC: Article writing. AC, MCM: Critical revision. All authors contributed to read and approved the version of the submitted manuscript.

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