

Paragonimiasis: First Case Reported in Brazil

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The authors present a case from a 59 years old white female Brazilian patient, based in Salvador-Bahia, Brazil's northeastern side area, who experienced irritative cough and progressive dyspnea, and, after 18 months, was admitted to a hospital with respiratory insufficiency. The physical exam showed diffuse rales in both hemithoraces. Initial leukogram showed 14,400 cells/ μ L with 14% of eosinophils and chest X-ray showed peribronchovascular infiltrate, predominating in the lower half of the lung fields, and small opaque nodules. The high-resolution computed tomography scan of the chest (HRCT) presented compatible pattern with airways disease, especially from the small airways, with air trapping, tree sprouting images, central lobular nodules and bronchiectasis, making the results compatible with bronchiolitis and bronchiectasis. The transbroncho biopsy unveiled granulomatous lesion with necrosis, where was noticed a structure compatible to a parasitic case, and the research of the parasite eggs in the sputum was positive to paragonimus. After the praziquantel use, the patient presented a thick ferruginous expectoration and the result for BAAR examination was positive. The PCR exam and the sputum culture confirmed *M. tuberculosis*, and then the treatment for *M. tuberculosis* was initiated. The authors warn that this infection may have been a consequence of economics globalization process, where the importation of parasitized crustaceans might be the cause. However, there is the need of an accurate examination for the possibility of paragonimus specimens in this area of Brazil.

Key-Words: Paragonimiasis, tuberculosis, respiratory insufficiency, bronchiolitis Brazil, case report.

Paragonimiasis is the result of an infection caused by one described type of paragonimus [1]. There are 50 described species of paragonimus and 10 of them are harmful to human beings. The most common is the *Paragonimus westermani*, which is endemic in China, Japan, Philippines and Korea. Other variety have been responsible for the contamination of human beings on the west side of Africa, South America and Central America [2]. Infections caused by *P. kellicotti* have been autochthonous within the United States of America [3], but most of the cases had been diagnosed on immigrants. No case was reported in Brazil so far. Most of the time, the contamination happens when the raw or undercooked crustaceans (crab or crayfish) infested by paragonimus on metacercaria stage are ingested. Eggs eliminated on the sputum or on human stools, in fresh water, after a while, release miracidia, which move freely through the water and contaminate the water snails. Inside them, the miracidia develop into cercariae and, once eliminated, they penetrate on the crustacean's muscle and then unfold to metacercariae. The social habit of ingesting raw food makes the ingestion of contaminated crustaceans the main cause of this disease. Other ways of contamination are: eating raw meat of mammal animals (dog, cat), which are also paragonimus final hosts; and through

contaminated utensils [2]. The metacercariae, in human duodeno, are released from the cysts and penetrate into the gastrointestinal wall, moving into the peritoneal cavity. Even though there is the chance of migration to the extra-lung sites, the majority of young paragonimus penetrate in the diaphragm and get to the pleural cavity and to lung parenchyma, where the young paragonimus may cause bronchopneumonia, interstitial pneumonia and bronchiectasis [4]. Around the parasite, emerges a group of eosinophils and neutrophils, and later on mononuclear cells. Necrosis of pulmonary parenchyma followed by the development of a fiber capsule surrounding the adult paragonimus happens in a next stage. By the seventh or eighth week, the eggs production starts inside the capsule, which size increases and might break often inside the bronchiolus. The expectorated material has parasite eggs, Charcot-Leyden's crystals, inflammatory cells and necrotized tissue.

The presence of eggs on the tissue leads to the development of granulomatous reaction. When the cysts do not have more viable parasites or are empty, they draw back and a scar may take place and become calcified [5]. Pleural effusion with empyema formation may appear with the parasite migration to the pleural cavity [6]. The disease can be spread to other areas of the organism [7].

The diagnosis is given finding parasites eggs in biologic specimens and/or the parasite identification in a histopathologic exam. Molecular biology techniques have been developed [8]. Serologic methods and ELISA have been used with sensibility and specificity of, respectively, 92% and 90%; nevertheless, the values remain high after starting the treatment [9]. The treatment of choice is the praziquantel [10].

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Case Report

SSP, 59 years old, white female Brazilian patient, married, teacher, based in Salvador, Bahia, northeastern side of Brazil. The irritative low-productive cough started in July 1998 associated with progressive dyspnea. Eighteen months afterwards, the patient was dyspneic even when resting, when she was then hospitalized. It was reported that, within the 2 years before the disease had showed up, the patient traveled twice to overseas – to Chile and Spain – and reported that she had eaten grilled salmon. The patient used to eat paella, a typical Spanish meal, that is made of seafood and crustaceous specimens. The physical exam presented diffuse rales in both hemithoraces. The result of the first leukogram showed 14,400 cells/ μ L with 14% of eosinophils and the chest x-ray showed peribronchovascular infiltrate, predominating in lower half lung fields, and small opaque nodules (Figure 1). The high-resolution computed tomography scan of the chest (HRCT) presented compatible pattern of airways disease, especially from small airways, with air trapping, tree sprouting images, central lobular nodules and bronchiectasis, making the results compatible with bronchiolitis and bronchiectasis (Figure 2). The initial spirometry revealed VC of 75% of what was expected and normal FEV₁. After 18 months, the result of the VC was 42% of what was expected.

A treatment with corticoid took action and a transbroncho biopsy revealed cartilaginous bronchus and granulomatous lesion with necrosis, where was observed a structure similar to a parasite capsule (Figures 3 and 4). An exam made with the sputum parasites eggs was indicative for paragonimus, and then sustained by “Center for Control Diseases –USA” (Figure 5).

Praziquantel was used in the treatment. After the use, the patient presented thick ferruginous expectoration, where paragonimus eggs were found again and the BAAR had a positive result. Dyspnea became worse, so the patient was hospitalized. After examination, the patient presented regular general state, no fever, cyanosis with respiratory frequency of 46 bpm, heart rate of 120 bpm, artery pressure of 110 x 80 mmHg and there were rales in the chest auscultation of both hemithoraces. At this time, the O₂ pulse saturation was 84 mmHg. The treatment for *M. tuberculosis* and *MAC complex* was initiated, and the treatment with praziquantel was repeated. The PCR exam and sputum culture were positive for *M. tuberculosis*, and the treatment for *MAC-complex* was then interrupted.

The patient presented a progressive recover. Nowadays the respiratory frequency is 20 bpm and the pulse saturation is 94% when resting, decreasing to 91% after a 6 minutes walk. The last spirometry revealed VC of 52% of what was expected and the chest HRCT presented subtotal retrocession of the lesions, but it was also noticed the remaining of shortness of breath, observed during expiration (Figures 6 and 7).

Discussion

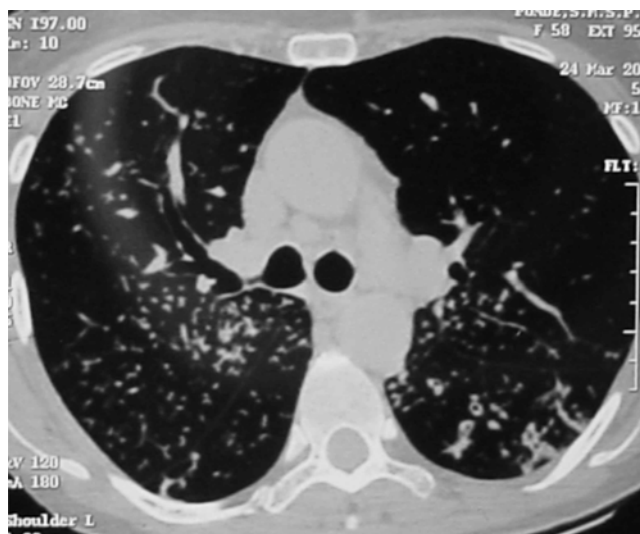
It is related to be the first case of paragonimiasis in Brazil. In South America, the majority of paragonimiasis reports had

been in Ecuador, with an annual incidence of 13.2 cases [11]. The clinic manifestations of cough, ferruginous sputum (result of the cyst rupture) and discovery of nodules and bronchiectasis on the x-ray examination are the most frequent features [4], and all have been noticed on this current case. The association with *M. tuberculosis*, in endemic area, is described in up to 30%. More than half of the cases are initially treated as tuberculosis [5,12]. Bronchiectasis and small multiple nodules had been described in the chest HRCT scanning, with positive culture for *MAC complex* [13]. These notes justify the decision of, initially, treat for both mycobacteria. The accomplishment of PCR for *MAC complex* and *M.*

Figure 1. X-ray showed peribronchovascular infiltrate, predominating in lower half lung fields, and small opaque nodules.



Figure 2. The high-resolution computed tomography scan of the chest (HRCT) presented air trapping, tree sprouting images, central lobular nodules and bronchiectasis, indicating bronchiolitis and bronchiectasis.



Figures 3 and 4. Transbroncho biopsy revealed cartilaginous bronchus and granulomatous lesion with necrosis.

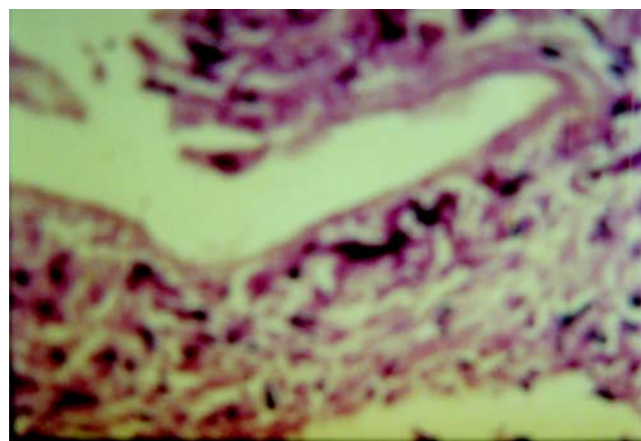
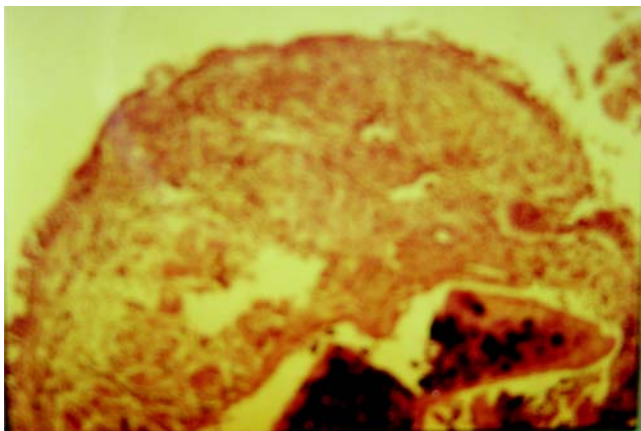
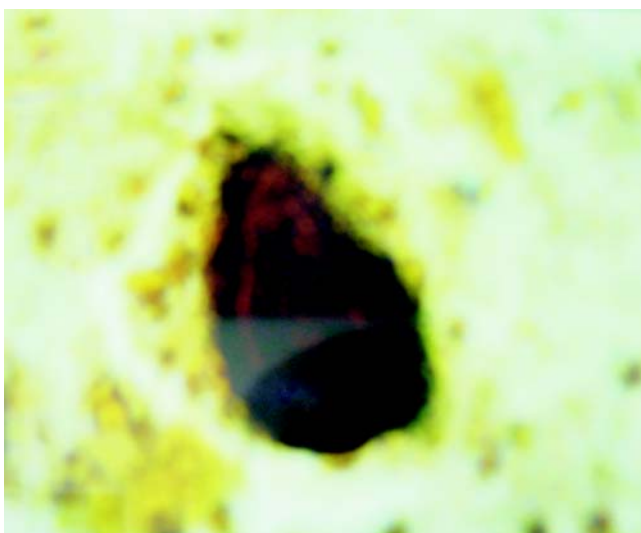


Figure 5. Parasite egg (paragonimus).

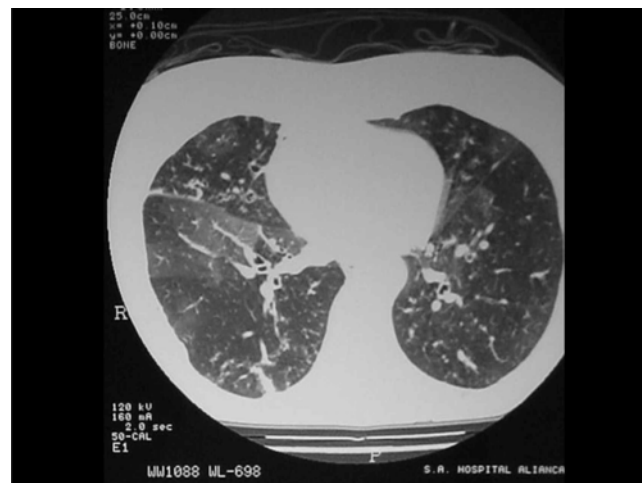


tuberculosis, should be indicated in this situation because it is a very fast exam.

However, the discovery of granulomatous bronchiolitis with caseous necrosis leading to respiratory insufficiency is not a very common finding caused by *M. tuberculosis* infection. Lemos et al related a case of granulomatous bronchiolitis with caseous necrosis due to *M. tuberculosis* that developed with breathing insufficiency, resulting in death, despite the treatment with tuberculostatic drugs [14]. On the authors' view, bronchiolitis on the current case was caused by paragonimus, and this was confirmed by transbroncho biopsy. The shortness of breath observed on the chest HRCT and the reduction of VC are more likely to be a consequence of a constrictive bronchiolitis, diffuse and residual, than a result of a peribronchiolar scar process after treatment.

Due to the fact that no cases of autochthonous paragonimiasis had been reported in Brazil, the authors concluded that the contamination could have happened as a consequence of the economic globalization process, where

Figures 6 and 7. Chest HRCT presented subtotal retrocession of the lesions. Remaining of shortness of breath, observed during expiration (Figure 7).



importation of parasitized crustaceans may be the cause. However, there is still necessity to evaluate this theory and even to verify if the coastal region of Bahia, northeastern side of Brazil, area with plenty of rivers and mangroves, where there are a large quantity of mollusks and crustaceans, and also where the patient always had lived, has or not paragonimus specimens.

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