Cláudio Dornas de Oliveira[1], Glaucio de Oliveira Nangino[2], Paulo César Correia[3], Carlos Vechio Salomão[3], Marcelo Alencar Resende[4], Leonardo da Cruz Peixoto[4], Maurício Buzelini Nunes[5]

Tumoral pulmonary mass secondary to *Schistosoma mansoni* infection resembling neoplasia: case report

Massa tumoral secundária a infecção por Schistosoma mansoni simulando neoplasia de pulmão: relato de caso

1. Physician of Santa Casa de Belo Horizonte – Belo Horizonte (MG), Brazil.
2. Physician of Santa Casa de Belo Horizonte – Belo Horizonte (MG), Brazil.
3. Physician of Santa Casa de Belo Horizonte – Belo Horizonte (MG), Brazil.
4. Physician of Santa Casa de Belo Horizonte – Belo Horizonte (MG), Brazil.
5. Physician of Santa Casa de Belo Horizonte – Belo Horizonte (MG), Brazil.

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Author for correspondence:
Cláudio Dornas de Oliveira
Santa Casa de Belo Horizonte- CTI adulto, 2º andar
Av. Francisco Sales, 1111- Santa Efigênia
CEP: 31150-221 - Belo Horizonte (MG), Brazil.
Fone: +55 (31) 3238-8181
E-mail: cdornas@yahoo.com.br

ABSTRACT

Patients with chronic *Schistosoma mansoni* infection may feature a range of pulmonary symptoms and radiological findings. Eggs, and rarely adult worms, may passively enter the pulmonary circulation, usually via the portal system, where they may cause pulmonary inflammation, fibrosis, hypertension and cor pulmonale. A 25-year-old patient who lived in a schistosomiasis endemic area with a pulmonary mass suggestive of malignancy underwent exploratory thoracotomy. The mass was adherent, with no resection possibility. The lung-biopsy specimen evaluation showed several granulomas with *Schistosoma mansoni* eggs and hyperplasic connective tissue with no sign of malignancy. The patient had respiratory failure and hypotension immediately post-surgery. Specific treatment (praziquantel) and prednisone were given. The patient had pneumonia and septic shock. The patient was given antibiotics, vasopressors, mechanical ventilation and hemodialysis with no improvement, and subsequently died 28 days after the surgery.

Keywords: Schistosomiasis; *Schistosoma mansoni*; Neoplasias/secondary; Lung diseases, parasitic; Case reports

INTRODUCTION

Patients with chronic schistosomiasis may have pulmonary involvement featuring a variable range symptoms and radiological findings.[1] The lungs may be involved due to anomalous eggs migration via portal system into the pulmonary artery system (via porto-systemic anastomosis) and less commonly by adult worms migration. There are extensive parenchyma involvement cases, as well as others with predominant arteritis, with pulmonary hypertension and *cor pulmonale.*

Lung parenchyma involvement is mainly characterized by granulomatous reaction to *Schistosoma mansoni* eggs. The granulomas have no preferential location, and may be found in all lung segments and pleura. Inter-alveolar thickening and connective tissue fibrosis were also reported.[2] Lesions from dead worms are rarer, and characterized by necrotic areas surrounded by intensive exudation, usually reabsorbed and involved by cicatricial tissue.[3,4]

A case involving a young female patient with atypical pulmonary schistosomiasis faking neoplasia is presented. Aspects regarding differential diagnosis are discussed.

CASE REPORT

This was a 25 years old female patient who lived in endemic schistosomiasis area and was referred to a thorax surgery service with dysphagia, weight loss and exertion dyspnea. There was no relevant history of previous diseases or comorbities. The physical examination evidenced severe malnutrition, asymmetrical thorax expansion and bronchial sounds at the lower left hemithorax. Additional tests showed: chest X-ray: heterogeneous hypo-transparency at lower left lobe (Figure 1); radiographic exam with contrast and upper digestive endoscopy: signs of esophageal lower third extrinsic compression; chest CT: heterogeneous mass taking the upper the left lower lobe basal lateral and basal posterior areas, with adjacent pleural thickening (Figure 2). Presence of two similar images, but smaller, on the upper lingula segment and anterior left upper lobe (Figures 3 and 4); increased left liver lobe (non-oriented examination); spirometry: moderate restrictive respiratory disorder; bronchoscopy: extrinsic left basal bronchia compression; bronchial biopsy: non-specific bronchitis.

Surgical approach was decided. Enteral nutrition support was started, as well as preoperative evaluations. The patient underwent exploratory thoracotomy. A large mass was found, invading the parietal pleura, left lung hilum, aorta, diaphragm and left atrium, with no resection possibility. A biopsy was performed and the patient was referred for post-operative follow-up at the intensive care unit. She required mechanical ventilation and vasoactive amines from the admission. She coursed with severe restrictive ventilatory disorder, and refractory hypoxemia. The histopathology revealed thickened and fibrosed pleura, pulmonary parenchyma with fibrosis and several granulomas containing *Schstosoma mansoni* eggs; no lesions suggesting neoplasia were found (Figure 5). The patient underwent praziquantel 50 mg/kg single dose treatment plus prednisone 1 mg/kg/day. Echo-Doppler and the Swan-Ganz catheter evidenced pulmonary hypertension. Lower limbs duplex scan was negative for venous thromboembolism.

Subsequently she developed pulmonary infection and

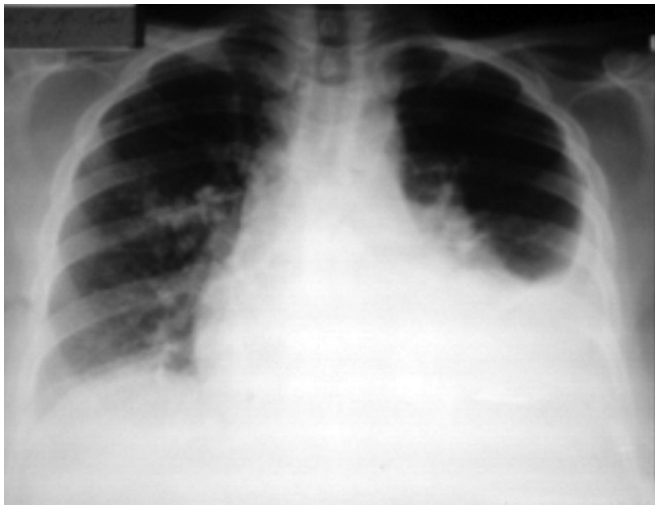

Figure 1 – Posterior-anterior chest X-ray: heterogeneous hypo-transparency at left lower lobe.

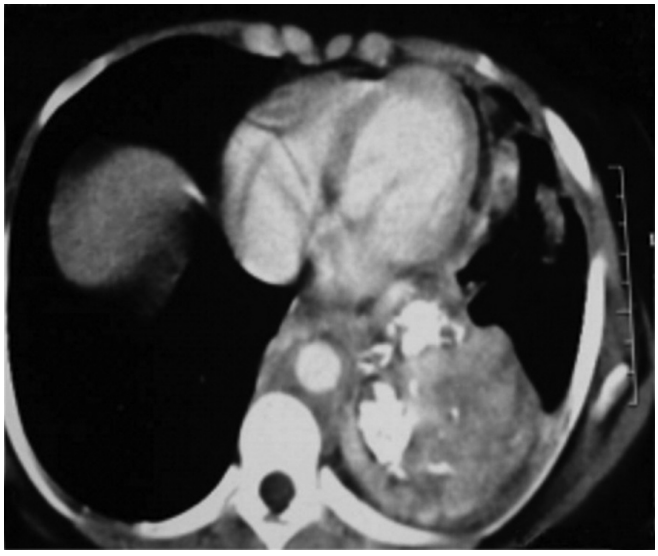

Figure 2 – Chest CT: heterogeneous mass at left lower lobe basal posterior part.

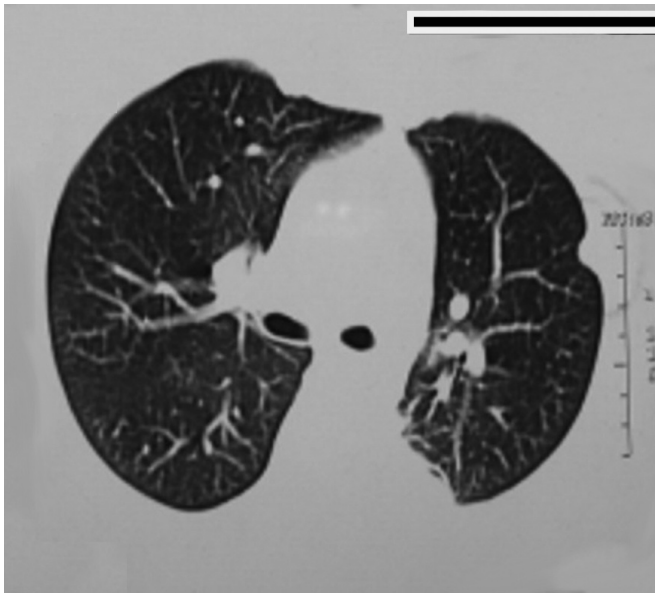

Figure 3 – Chest CT: mass at left lower lobe basal posterior portion.

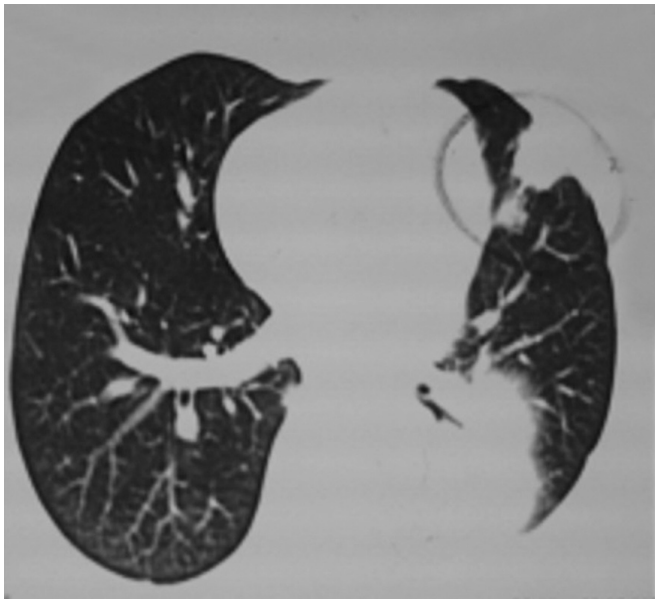

Figure 4 – Chest CT: nodular image at left upper lobe upper portion, adjacent to the pleura.

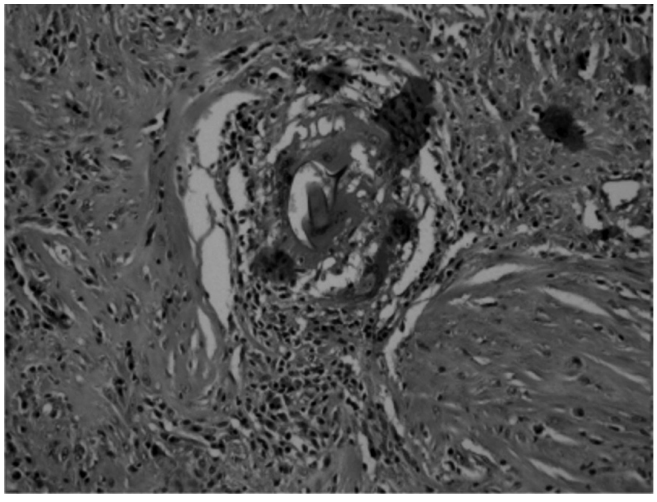

Figure 5 – Pathology – fragment of lung: schistosomotic granuloma, with inflammatory infiltrate surrounding the lesion, and connective hyperplasia.

septic shock. Antibiotics, vasoactive amines, ventilatory support and hemodyalisis, were given with no improvement. Subsequently she died, 28 days after the surgery.

DISCUSSION

From a radiological point of view, chronic schistosomiasis pulmonary parenchyma changes are commonly described as: diffused infiltrate, focal opacities and micro-nodules.[5] Pulmonary nodes secondary to schistosomiasis are rare, and pose a differential diagnosis with lung neoplasia, and are frequently defined only after exploratory thoracotomy.[6-8] The term 'pseudotumoral schistosomiasis presentation' was first used in 1975 describing a pulmonary node in a patient who died, being a autopsy findings granulomatous reaction, *Schistosoma mansoni* eggs, and pulmonary arterioles obliteration.[9]

The presence of a large mass associated to *Schistosoma* infection (bilharziasis) was described in 1953 in Cairo.[10] The gross presentation during the surgery suggested lung neoplasia, requiring pneumectomy. Similarly, the histology showed fibrotic and thickened pleural tissue with cicatricial granulomas, fibrosed lung tissue, *Schistosoma* eggs surrounded by histiocytes, eosinophils and fibroblasts. The authors pointed out that, as first hypothesis, the lung mass producing these pathology findings was secondary to schistosomiasis. The schistosomiasis pseudo-neoplasic forms represent anomalous response to the parasite eggs. Hyperplastic connective tissue formations develop around the granulomatous formations, as a host reaction.

It is proposed that, in schistosomiasis endemic areas, pulmonary schistosomiasis is considered a differential diagnosis for complex structures, as pulmonary masses.

RESUMO

Indivíduos infectados com *Schistosoma mansoni* na fase crônica da doença podem apresentar comprometimento pulmonar com sintomatologia e alterações radiológicas variáveis. Os pulmões podem ser acometidos pela migração anômala de ovos do sistema porta para o sistema arterial pulmonar (através de anastomoses porto-sistêmicas) e menos comumente por migrações ectópicas de vermes adultos. Há casos com extenso comprometimento parenquimatoso e outros com predomínio de arterites, com hipertensão pulmonar e cor pulmonale. Paciente jovem, residente em área endêmica de esquistossomose, com massa pulmonar sugestiva de neoplasia foi submetida a toracotomia exploradora sem possibilidade de ressecção da massa. Exame histopatológico mostrou vários granulomas esquistossomóticos e hiperplasia do tecido conjuntivo, sem sinais de neoplasia. Evoluiu com insuficiência respiratória e instabilidade hemodinâmica no pós-operatório imediato. Recebeu tratamento específico (praziquantel) associado a prednisona. A paciente cursou com infecção pulmonar e choque séptico. Recebeu antibioticoterapia, aminas vasoativas, suporte ventilatório e tratamento hemodiálitico sem melhora. Evoluiu para óbito 28 dias após cirurgia.

Descritores: Esquistossomose; *Schistosoma mansoni*; Neoplasias/secundário; Pneumopatias parasitárias; Relatos de casos

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