

## Case Report

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# A Case of Pulmonary Nodular Schistosomiasis

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Bilharzia is a parasitic infection particularly affecting the digestive tract and urinary tract. Lung involvement is rarely reported. We report a case of pulmonary bilharzioma of nodular type surrounded by ground glass opacities diagnosed on CT-scan and associated with a hepatic nodule, in a 41-year-old woman. The disappearance of the pulmonary nodule under antischistosomal treatment made it possible to make the diagnosis a posteriori without going through an invasive process.

**Key words:** Antibilharzien; CT scan; Ground glass opacities; Nodule; Pulmonary bilharzioma

## INTRODUCTION

Bilharzia is a parasitic infection endemic to 70 countries (1). Lung involvement would concern 15% of the infected population (1,2). Pulmonary schistosomiasis is rarely reported, although it can simulate primary or secondary tumor lesions. We report the case of a pulmonary schistosomiasis nodule discovered during the exploration of chronic coughs in a 41-year-old woman who simultaneously presented a hepatic nodule in order to describe its management.

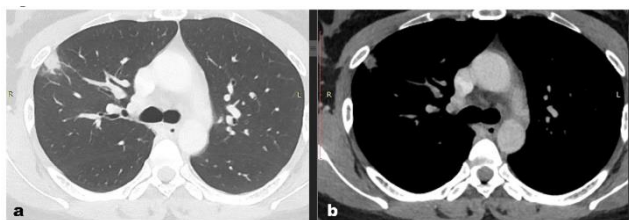
## CASE SUMMARIES

This was a 41-year-old woman, a bureaucrat in the western region of Madagascar, who presented episodes of chronic dry coughs which justified a consultation in the

Thoracic Surgery service. She had no particular history. She was not an active or passive smoker. She lived with two people in her home. No notion of chronic cough was reported among her relatives. She was ignorant of any notion of fever. Clinical examination did not show any significant abnormality. The general condition was maintained and there was no noticeable weight loss.

Schistosoma serology was positive and there was an eosinophilic polynuclear leukocytosis. The tests for eggs bilharzia in stool and urine were negative. Genexpert test for Koch bacillus DNA was negative. Hepatic transaminases, hepatitis and amoebic serology as well as gamma globulin transferase, bilirubinemia and alkaline phosphatase were unremarkable.

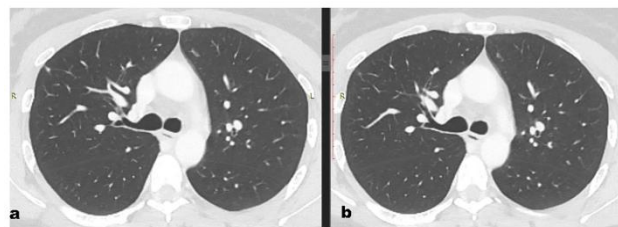
The frontal chest X-ray did not show any abnormal opacity. The persistence of the clinical symptomatology motivated the realization of a thoraco-abdominal CT scan which showed a solid nodule under the right upper lobar pleural area measuring 15 x 14 mm, surrounded by ground glass opacity, facing the anterior arch of the fourth rib (Figure 1). This nodule did not show significant contrast enhancement. There was also a hepatic subcapsular low density nodule, with peripheral enhancement in arterial phase and homogenizing in portal and late phase in segment VI. Supplement ultrasound revealed a homogeneous and well-limited hyperechoic aspect of this nodule.



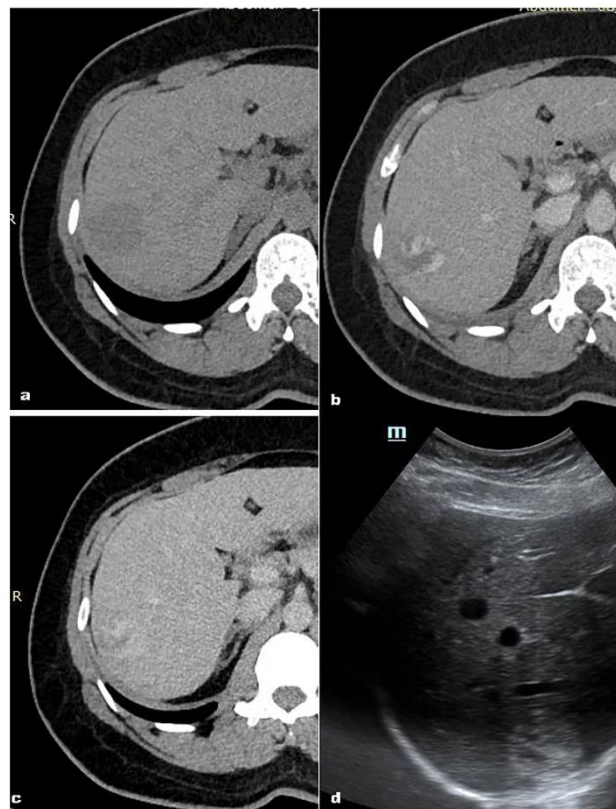
**Figure 1.** Axial CT scan images of the thorax, in parenchymal window (a) and in mediastinal window, with contrast media (b) showing a solid nodule surrounded by an area of ground glass in the right apico-ventral subpleural area, behind a costal arch.

In view of the atypical appearance and small size of the nodule and the positivity of bilharzia serology, awaiting further check, an anti-bilharzia treatment test with praziquantel at 40 mg / kg in a single dose was initiated. Three months later, control of bilharzia serology returned to normal, and CT-scan showed reduction of more than 75% in the size of the nodule and disappearance of the surrounding ground glass opacity (Figure 2a). The third CT scan performed three months from the previous one showed the disappearance of the nodular lesion (Figure 2b). The hepatic subcapsular nodular lesion was stable in size and density on the three examinations (Figure 3). There was no pulmonary artery abnormality on the three scans performed.

The cough improved gradually to disappear by the third month of follow-up. No other respiratory clinical manifestation occurred at 12 months follow-up.



**Figure 2.** Axial CT scan images of the thorax at three months of antischistosomal treatment (a) showing the marked reduction in the size of the nodule and the disappearance of the ground glass areas, and at six months of treatment (b) showing the disappearance of the nodule.



**Figure 3.** Abdominal CT scan images passing through the liver, showing a low density subcapsular nodule (a), with peripheral and centripetal enhancement in arterial phase (b) and homogenization in portal phase (c). Sagittal ultrasound section of the right liver passing through segment VI showing the hyperechoic aspect of the nodule (d).

## DISCUSSION

Bilharzia is an endemic parasitic infection in 70 countries with approximately 200,000 infestations per year (1,2). Madagascar is home to two types: intestinal bilharzia on the eastern seaboard and urinary schistosomiasis in the western part. Both types can reach the lung by haematogenous migration of eggs along the pulmonary

arteries. While other species bilharzia have poor lung migration capacity, eggs of *Schistosoma haematobium* laid in perivesical plexuses and the inferior vena cava migrate to the lungs more than those of *Schistosoma mansoni* located in the liver.

Acute schistosomiasis, known as Katayama syndrome, which manifests as acute fever, dry coughs and malaise, usually presents between the 3rd and 8th week of infestation in people who have never had contact with the parasite or in the event of a massive infestation and can heal itself (3,4). It is due to the larval migration responsible for an immuno-allergic reaction. Other symptoms such as headache, myalgia or even dyspnea and hemoptysis as well as signs of meningoencephalitic involvement may occur. During this phase, there is a moderate biological inflammatory syndrome with high CRP and serum amyloid A; slight acceleration in red blood cell sedimentation rate, eosinophilic leukocytosis (3).

Thromboembolic phenomena are accompanied by larval migration in the arterioles but also the presence of eggs surrounded by an inflammatory granulomatous reaction in the tissues (4). In the lungs, granulomatous reactions will cause pulmonary arterial hypertension either by arterial migration of the eggs or secondary portal hypertension linked to hepatic damage.

The diagnosis of schistosomiasis is based on schistosoma serology associated with hypereosinophilia and arises when there is evidence of eggs of bilharzia in the stool and / or urine (5). The notion of contact or bathing in stagnant fresh water in an endemic area should be researched. Pulmonary involvement is facilitated by cirrhosis, with the development of collateral venous circulation, frequent in patients with bilharzia in endemic areas (6) even if most of the cases of pulmonary schistosomiasis reported concern travelers and natives of non-endemic areas.

The chest X-ray may be abnormal in the acute phase. In seven of a series of 10 patients, Foti et al. (5) found radiographic lesions while the CT scan was abnormal for all 10 patients. The lesions are nodular in five patients,

consolidation in three patients and ground glass in four patients. Foti's series (5) shows the sensitivity of the scanner compared to the radiography with a total of 15 nodules and four ground glass lesions detected against 91 nodules and eight ground glass lesions on the scanner even though these two techniques detected as many consolidation lesions. Indeed, lesions of small size, or made of ground glass alone or lesions of medium size but with a prevalent peripheral halo may not be visible on radiography. In a series of 10 patients, the lesions measure between 2 to 5 mm in five patients and 5 to 15 mm in four patients in the study by Nguyen et al. (7), while Rakotovo et al. (8) reported a pseudotumoral form with a size of about 5 cm. The lesions have no predilection for apical or basal, peripheral or central involvement. The nodular lesion was surrounded by a ground glass surface and measured 15 mm long axis in our patient and was located in the right upper lobe; its non-visualization on the X-ray could be explained by its small size but also its location under the pleura opposite a costal arch. This costal interposition on the lesion would also limit a transparietal biopsy under radiological guidance. The ground glass opacities are nonspecific and do not correlate with either the extent of the infestation, the eosinophilia level or the size of the nodule (7). In addition, Soares Souza et al. (9) reported an atypical form producing a diffuse and bilateral infiltrative reticulonodular disease confirmed by pulmonary biopsy after thoracotomy. This process seems to be too invasive if the diagnosis can be made by a therapeutic test with reduction or disappearance of the lesions under antischistosomal directed by clinic and biology aspects. In our patient, the reduction of the lesion was observed on the CT scan at three months and disappearance at 6 months, thus making it possible to avoid surgical or transparietal biopsy. The association in our patient with a hepatic nodule suggested a secondary tumor process even though the hepatic lesion presented characteristics compatible with a hemangioma. Indeed, the lesion is spontaneously homogeneous and low density with peripheral enhancement in the arterial phase and

homogenization in the portal phase and that on ultrasound it presented a homogeneous hyperechoic aspect. The three-month and six-month controls did not show any change in the size or in the vascular kinetics of this lesion, while the lung lesion regressed and then disappeared, testifying thus to a pure chance of this association. On the other hand, Lagler et al. (3) reported multiple bilateral pulmonary nodules and hepatic low density micronodules in a bilharzian patient, which they linked either to foci of abscesses or secondary lesions but the disappearance of the lesions after antischistosomal treatment made it possible to make the diagnosis a posteriori. The fear of these nodular lesions or pulmonary masses is cancer, which would explain the adoption of an invasive diagnostic process by some authors. Thus, the therapeutic test must be associated with close monitoring to avoid delay in the management of a possible tumor (5). With these nodules, thin-walled cystic lesions can coexist in the acute phase of the infection, as in two of the ten cases reviewed by Foti et al. (5).

In the chronic phase, the pulmonary lesions of bilharzia are characterized by sequelae of granulomas which may gradually calcify but above all by vascular damage such as pulmonary arterial hypertension linked to the sequelae of focal lesions of granulomas or to liver damage realizing porto-pulmonary syndrome. Experimental study carried out on mice demonstrates the relationship between these granulomatous lesions and vascular remodelling (10). In fact, vascular lesions are characterized by a muscular thickening of the wall of the pulmonary arteries of medium and large caliber near the schistosome granulomas. Thus, the lesions are of non-uniform distribution in the lungs and therefore depend on the location of the areas where the parasite eggs are deposited around which the granuloma will develop. These vascular lesions remain rare, however, since Foti et al. (5) did not detect any on the tomodensitometry measurements of the diameter of the pulmonary arterial trunk in ten subjects suffering from chronic schistosomiasis, whereas all these subjects presented pulmonary lesions either of consolidation type

or nodule (solid or mixed), ground glass or even hilar adenomegaly.

The single dose of 40 mg / kg of Praziquantel allows, as in our patient, the disappearance of the images and the return to normal of the schistosome serology. The disappearance of the CT images is on average 35 days after treatment on the Foti et al. series (5). This rather short time allows to make up for a possible alternative diagnosis on the CT scan, in particular on large lesions for which the observation of reduction or disappearance of the lesion makes it possible to avoid an invasive attitude.

## CONCLUSION

Pulmonary schistosomiasis can simulate tumors especially if they are associated with other lesions such as liver nodules. Comparison of the clinical data with the radiological images makes it possible to propose a therapeutic test and to evaluate the lesions early in order to avoid an aggressive strategy.

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