Case Report

Pleural schistosomiasis masquerading as tubercular pyopneumothorax: World's first case report

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ABSTRACT

Schistosomiasis is an uncommon waterborne helminthic infection that infects humans. Although it is not prevalent in India, many cases are reported due to contact with infested water bodies. Schistosomiasis primarily involves the lower urinary tract and colorectal region. Pulmonary schistosomiasis, although very unusual, has been described with the systemic manifestation of the disease; however, pleural involvement with schistosomiasis has never been described before in the literature. We report this first case of pleural schistosomiasis masquerading as tuberculosis, which posed a diagnostic challenge and later a new learning point in the presentation of the disease.

KEY WORDS: Helminthic, pyopneumothorax, schistosomiasis, tuberculosis, waterborne

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INTRODUCTION

Schistosomiasis is a water-bourne helminthic infection which usually affects the lower urinary tract and colorectal region. Pulmonary schistosomiasis although very unusual, has been described with the systemic manifestation of the disease; however, pleural involvement with schistosomiasis has never been described before in the literature. The authors report this rare case of pleural schistosomiasis masquerading as tuberculosis which posed a diagnostic challenge.

CASE REPORT

A 26-year-old female, college student, nonsmoker resident of Assam, had a history of dry cough along with high-grade intermittent fever for the past 1 month. She complained of nonradiating right-sided chest pain with progressive breathlessness, modified Medical Research Council (mMRC) Grade 1 to mMRC Grade 3, for the last 15 days. She had lost 8 kilograms of weight in the last 1 month. She was evaluated initially at outside hospital where chest X-ray done showed air–fluid level in the right hemithorax [Figure 1]. Computed tomography (CT) of the chest was done, which showed moderate hydropneumothorax with thickening of overlying pleura. Collapse consolidation of the right lower lobe was noted and the remaining lung parenchyma and pulmonary vasculature were normal. There was no significant mediastinal lymphadenopathy.

Right intercostal drainage (ICD) with water seal was placed and the pleural fluid analysis showed frank pus with glucose 20 mg/dl, total protein 6.14 g/dl, albumin 2.74 g/dl, total leukocyte count (TLC) count 7842/mm^3

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with polymorph predominance, lactic acid dehydrogenase 2183 U/L, and adenosine deaminase (ADA) 119.7 U/L. Pleural fluid cytology showed acute inflammatory process with no evidence of malignant cells. The patient was started on antitubercular treatment (comprising four drugs: rifampicin, isoniazid, pyrazinamide, and ethambutol). She continued to have persistent air leak on ICD with continuous pleural fluid drainage of more than 100 ml daily. She did not tolerate the medications due to severe gastritis, and the patient was then referred to our facility for further management with ICD *in situ*. She was afebrile on admission and was hemodynamically stable with blood pressure of 110/70 mmHg and oxygen saturation of 95% at room air. General physical examination showed no pallor, icterus, clubbing, or pedal edema. JVP was not raised and neck veins not engorged. Cardiovascular system examination revealed tachycardia with no murmurs. Respiratory examination showed amphoric breath sounds on the right hemithorax with ICD *in situ* with a continuous air leak (Grade 4). Her routine laboratory investigations were normal, except for anemia and raised peripheral eosinophilia (hemoglobin of 9.6 mg/dl and normal TLC with eosinophilia 552/mm^3); viral markers were negative. Electrocardiogram and two-dimensional echocardiography were reported normal. Abdomen ultrasonography revealed a well-defined echogenic lesion of size 5.7 cm × 4.4 cm in the right lobe of the liver likely to be hemangioma. Stool routine microscopy was normal and serology for amoebic antibodies was negative.

Thoracic surgeon opinion was taken in view of persistent air leak and nonexpanding lung with pyopneumothorax, and the patient underwent right-sided video-assisted thoracic surgery (VATS)-assisted decortication along with the closure of bronchopleural fistula (BPF). Thickened pleura was decorticated and the examination of the tissue sent showed no acid-fast bacilli on smear and no pyogenic growth; GenXpert for mycobacterium tuberculosis was negative. Histopathology of the pleural tissue showed necrotizing granuloma, but on careful search revealed refractile oval parasitic eggs of Schistosoma clinching the diagnosis [Figure 2]. Enzyme-linked immunosorbent assay for specific serum immunoglobulin (IgG) antibodies was positive, but IgG Western blot was negative for serology. The patient was treated with praziquantel and she responded well to the treatment and as discharged after complete recovery. The patient remained asymptomatic at 6 months of follow-up.

DISCUSSION

Schistosomiasis also known as Bilharziasis is an uncommon parasitic infection caused by Schistosoma species. It is considered as nonendemic in India; however, many cases have been reported due to contact with contaminated water with human and animal feces.[1] Pulmonary schistosomiasis is an unusual occurrence as lungs are not the primary target organ to be involved. Although common parasitic pleural involvement is reported with amoebiasis, echinococcosis, and paragonimiasis, pleural schistosomiasis is not a known entity; our case presented with primary pleural involvement mimicking bacterial infection, which is the first case to be described in literature.

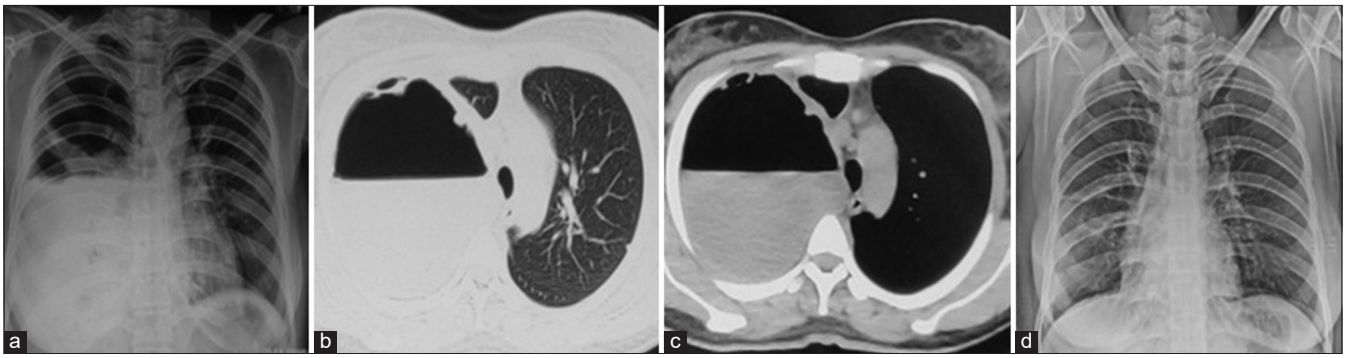

Figure 1: (a) Chest X-ray showing air–fluid level – right hemithorax. (b and c) Computed tomography images showing air–fluid level. (d) Chest X-ray after 1-month follow-up

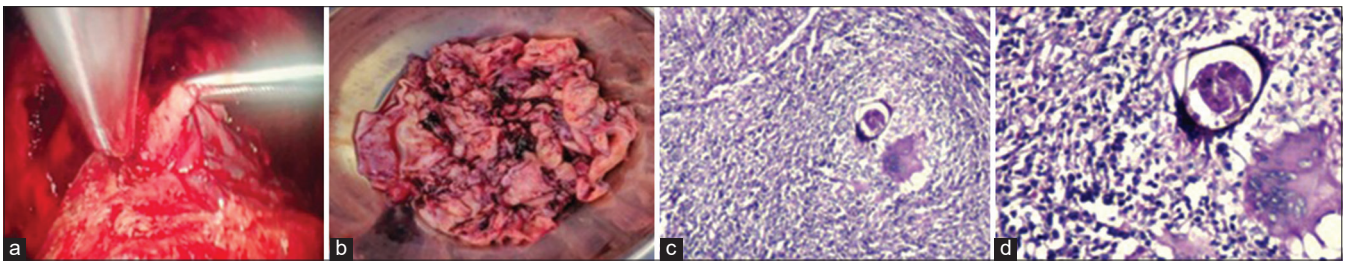

Figure 2: (a and b) Video assisted thoracic surgery decortication and repair of fistula apical segment of the right lower lobe. (c and d) Necrotizing granuloma with refractile oval parasitic egg of Schistosoma

Infection with schistosome occurs when people contact fresh water bodies infested with larvae cercariae released by specific intermediate host snails that have been previously infected by miracidia released from the eggs.[2] These larval forms of parasite reproduce in the intestine, liver, kidneys, and blood vessels, which get trapped within the target organ resulting in clinical manifestation.[3]

Pulmonary manifestations of the disease include acute and chronic disease. Acute form of disease is immune complex mediated usually presents as dry cough, shortness of breath, fever, weight loss, and eosinophilia within 3–8 weeks after exposure. The chronic form occurs months to years after exposure which leads to granuloma formation, vascular remodelling, pulmonary hypertension and cor pulmonale.[4] Our patient had acute pulmonary symptoms before presenting the medical facility and laboratory investigations showed peripheral eosinophilia.

Chest imaging of the lung can be normal or show discrete nodules, diffuse ground-glass opacities, and even mass-like lesion that mimic tuberculosis or malignancy. Chronic disease can show cardiomegaly and pulmonary artery enlargement.[5] There have been few case reports where the presentation of pulmonary schistosomiasis resembled that of pulmonary tuberculosis.[6]

The diagnosis is based on the demonstration of schistosome egg in urine or stool by direct microscopy or biopsy of the tissue involved. Serology tests are useful in acute cases, but cannot differentiate between current or past infection. Histopathology of the decorticated pleura in our case showed the presence of necrotizing granuloma with refractile Schistosoma egg. Species identification could not be done in our case; however, *Schistosoma haematobium* has been reported commonly in India in several studies.[7] The use of bronchoscopy or lung biopsy may be considered. Indirect evidence of infection may also be found by demonstrating hepatosplenic or genitourinary schistosomiasis.[8] Treatment for schistosomiasis infection is praziquantel, which has been shown to be effective in acute and chronic forms of the disease.[9]

Our case posed a diagnostic challenge due to difficulties involved in differential diagnosis. The possibility of a bacterial infection and higher possibility of tuberculosis due to pyopneumothorax and elevated ADA levels was initially considered. Tuberculosis remains the most common cause for the same due to higher prevalence in India, but emphasizes need to raise suspicion for alternate diagnosis in very high pleural fluid ADA. VATS decortication and repair of the BPF was done with confirmation of the diagnosis reinforcing the fact that early thoracic intervention is crucial in such challenging cases. The patient did improve promptly with treatment and remained asymptomatic thereafter. This case highlights the importance of multidisciplinary approach in pyopneumothorax with persistent air leak, which was misdiagnosed initially, but was found to be pleural schistosomiasis, leading to appropriate management and complete recovery.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Agrawal MC, Rao VG. Indian schistosomes: A need for further investigations. J Parasitol Res 2011;2011:250868.
2. Grimes JE, Croll D, Harrison WE, Utzinger J, Freeman MC, Templeton MR. The roles of water, sanitation and hygiene in reducing schistosomiasis: A review. Parasit Vectors 2015;8:156.
3. Verjee MA. Schistosomiasis: Still a cause of significant morbidity and mortality. Res Rep Trop Med 2019;10:153-63.
4. Schwartz E. Pulmonary schistosomiasis. Clin Chest Med 2002;23:433-43.
5. Martínez S, Restrepo CS, Carrillo JA, Betancourt SL, Franquet T, Varón C, *et al*. Thoracic manifestations of tropical parasitic infections: A pictorial review. Radiographics 2005;25:135-55.
6. Schaberg T, Rahn W, Racz P, Lode H. Pulmonary schistosomiasis resembling acute pulmonary tuberculosis. Eur Respir J 1991;4:1023-6.
7. Kali A. Schistosome infections: An Indian perspective. J Clin Diagn Res 2015;9:DE01-4.
8. Morris W, Knauer CM. Cardiopulmonary manifestations of schistosomiasis. Semin Respir Infect 1997;12:159-70.
9. Li YS, Sleigh AC, Li Y, Tanner M, Dessein A, Williams GM, *et al*. Five-year impact of repeated praziquantel treatment on subclinical morbidity due to Schistosoma japonicum in China. Transactions of the Royal Society of Tropical Medicine and Hygiene. 2002;96:438-43.