Case Report

An Unusual Cause of Bilateral Pneumothoraces

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Abstract

Pulmonary strongyloidiasis is an unusual manifestation of *Strongyloides stercoralis* infection typically seen in the immunocompromised patients. We report a case of *Strongyloides* hyperinfection in a patient with chronic obstructive pulmonary disease (COPD) who presented with bilateral pneumothoraces and pneumomediastinum. This is a rare presentation of the parasitic infection. An incidental finding of *Strongyloides* larvae in the respiratory secretions unexpectedly clinched the diagnosis and helped manage the patient optimally. **[Indian J Chest Dis Allied Sci 2020;62:153-156]**

Key words: Pulmonary strongyloidiasis, Hyperinfection syndrome, Bilateral pneumothorax, Pneumomediastinum

Introduction

Strongyloides stercoralis is a human intestinal nematode, endemic in tropical and subtropical countries including India. Most infections remain asymptomatic; however, severe strongyloidiasis such as hyperinfection and disseminated disease can occur in immunocompromised hosts. We report a case of *Strongyloides* hyperinfection in a chronic obstructive pulmonary disease (COPD) patient with bilateral pnuemothoraces and pneumomediastinum.

Case Report

A 52-year-old male presented with acute worsening of dyspnoea and right-sided chest pain for six hours prior to admission. There was no history of fever, increase in cough or sputum production. He was a reformed smoker and was diagnosed with COPD four years ago for which he was taking inhaled bronchodilators intermittently along with short courses of over-the-counter oral corticosteroids for exacerbations. He had no other co-morbidities.

On examination, he was hypoxic with an oxygen saturation of 82% on room air with tachypnoea. Examination of the respiratory system was suggestive of a right-sided pneumothorax with extensive rhonchi in the left hemithorax. With supplemental oxygen, the saturation improved to 92%. Arterial blood gas analysis showed hypoxaemic respiratory failure. Chest radiograph confirmed a right-sided pneumothorax (Figure 1A). An intercostal drain (ICD) was inserted and he was started on systemic steroids and inhaled bronchodilators. Post-ICD insertion, his dyspnoea and oxygenation improved. Complete blood count showed leucocytosis with neutrophilia (Table) and he was started on broad spectrum-antibiotics. Renal and liver function parameters were unremarkable and human immunodeficiency virus (HIV) serology was negative.

After approximately four hours of the above treatment, breathlessness worsened and he developed extensive subcutaneous emphysema involving the face, thorax, upper limbs and abdomen. The ICD position was re-checked; there was no evident leak at the insertion site and fluid column movement was present. A left-sided pneumothorax was suspected; but clinical examination was limited by the extensive subcutaneous emphysema. Blood gas analysis showed worsening of hypoxia and hypercapnia. In view of worsening respiratory distress, he was intubated and shifted to the intensive care unit and invasive mechanical ventilation was instituted.

On re-evaluation, clinical examination was suggestive of a left-sided pneumothorax and an ICD was placed on the left side pending any radiological investigation given the emergent situation (Figure 1B). Computed tomography of chest showed evidence of bilateral residual pneumothoraces with pneumomediastinum and bullous emphysema of the both lungs (Figure 2).

Culture of the endotracheal aspirate did not grow any organism but was observed on Gram staining motile larvae of *Strongyloides stercoralis* (Figure 3A). Thereafter, a stool examination was done which also revealed *Strongyloides* larvae (Figure 3B). He was started on oral albendazole (400mg once daily) and ivermectin (15mg once daily). He was successfully weaned off after two days and extubated. The patient denied any abdominal symptoms or skin eruptions. He showed steady improvement clinically and radiologically; the

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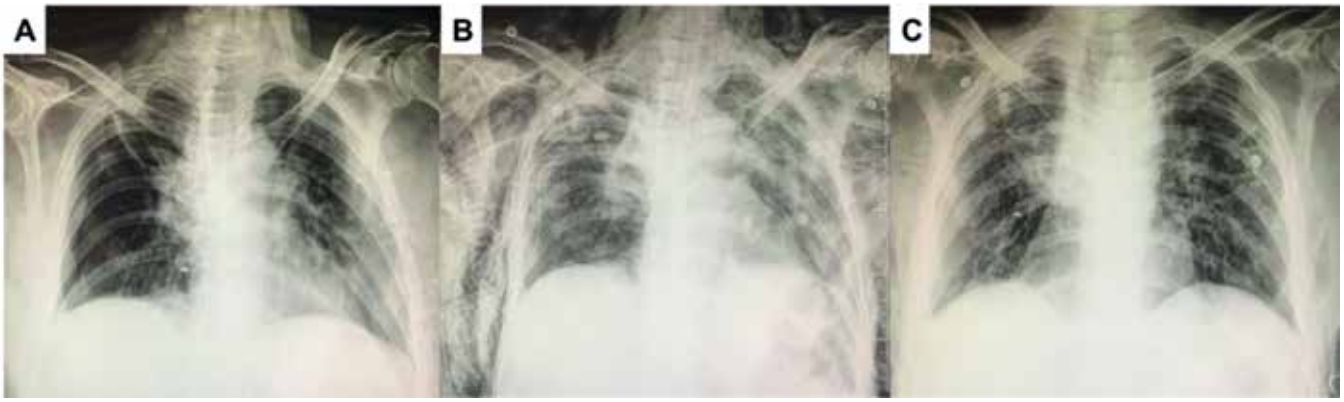

Figure 1. Chest radiographs (postero-anterior view): (A) *at admission* showing pneumothorax on the right side, (B) *Day 2*– showing subcutaneous emphysema with right ICD *in situ* and (C) *at discharge* showing resolution of pneumothorax and subcutaneous emphysema.

Table. Laboratory parameters of the patient before intubation

Parameter	Value
Hemoglobin (g/dL)	13.4
Total leucocyte count (cells/mm^3)	24850
Differential count	N91L4E0
Platelet count (cells/mm^3)	287000
Urea/ Creatinine (mg/dL)	24/ 0.8
Total bilirubin (mg/dL)	0.6
SGOT/ SGPT (U/L)	24/ 20
Serum albumin (g/dL)	3.2
Arterial blood gas (at admission)	
pH	7.45
PO_2	45mmHg
PCO_2	32mmHg
HCO3	30mmol/L
Arterial blood gas (after 4 hours; prior to intubation)	
pH	7.36
PO_2	42mmHg
PCO_2	48mmHg
HCO3	32mmol/L

Definition of abbreviations: SGOT=Serum glutamic-oxaloacetic transaminase; SGPT=Serum glutamic-pyruvic transaminase; PO_2=Partial pressure of oxygen; PCO_2=Partial pressure of carbon dioxide; HCO3=Bicarbonate

intercostal drains were removed sequentially after povidone-iodine pleurodesis (Figure 1C). Repeat sputum and stool examinations were negative for any ova or larvae. Ivermectin was continued for two weeks. Patient was discharged on domiciliary oxygen therapy in view of hypoxaemia.

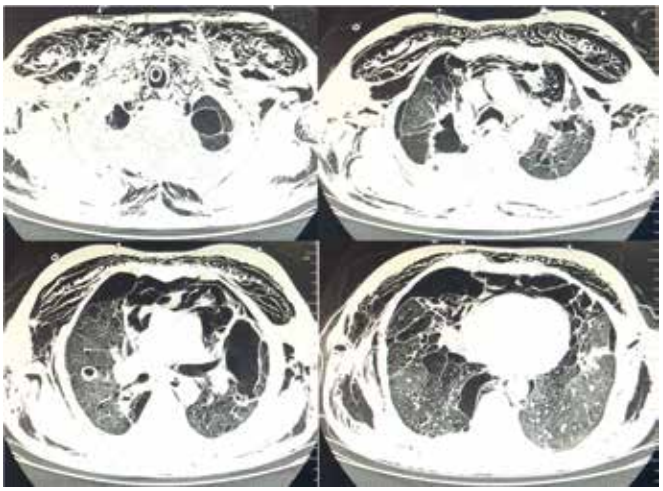

Figure 2. Computed tomography of the chest showing extensive subcutaneous emphysema with bilateral pneumothoraces with pneumomediastinum; lungs show evidence of bullous emphysema.

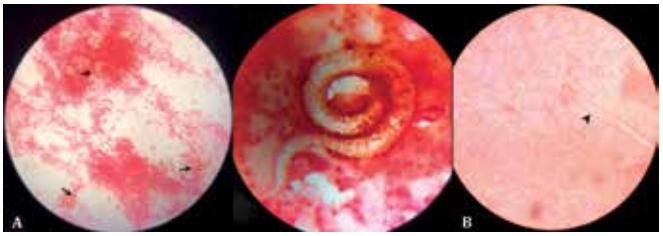

Figure 3. Bronchoalveolar lavage showing multiple larvae (arrows) of *Strongyloides stercoralis* in low power (left) and high power (right) and (B) stool wet mount showing motile larvae of *Strongyloides stercoralis* (arrow-head).

Discussion

Strongyloidiasis is an infection caused by the nematode *Strongyloides stercoralis*. This nematode is endemic to the tropical and sub-tropical regions. The parasites from infected soil penetrate the skin and reach the lungs,

where these traverse the pulmonary capillaries and enter the alveoli. From there these ascend in the trachea, are swallowed and reach the intestines where the adult worm develops and eggs hatch into rhabditiform larvae which are excreted. Occasionally, especially in the immunocompromised patients, the rhabditiform larvae develop into infective filariform larvae within the intestinal tract and penetrate the intestinal wall or the peri-anal skin to restart the life cycle – referred to as autoinfection.[1] The term hyperinfection syndrome refers to an accelerated autoinfection limited to the organs normally involved in the life cycle (intestines and lungs). When the parasite migrates to organs other than the lungs and the intestines, it is referred to as disseminated strongyloidiasis.[2]

The most common risk factors for hyperinfection syndrome and disseminated strongyloidiasis include corticosteroid therapy, drug-induced immunosuppression, achlorhydria, chronic kidney disease, HIV and human T-cell lymphotropic virus-type-1 (HTLV-1) infection and malnutrition. Chronic lung diseases may also predispose, possibly due to frequent use of steroids and a postulated delay in the transit of larvae due to fibrosis and architectural distortion leading to maturation of larvae in the lungs.[3]

The spectrum of pulmonary manifestations ranges from asymptomatic eosinophilia and exacerbation of asthma to fatal respiratory failure, alveolar haemorrhage and acute respiratory distress syndrome. Chest radiograph findings range from focal or diffuse infiltrates, consolidation and pleural effusion. Computed tomography findings include focal and diffuse ground-glass opacities, pleural effusion and thickened interlobular septae.[4] The occurrence of bilateral pneumothoraces has been reported only once in the literature.[5] In a similar case report in a patient with ascariasis, it was postulated that the pneumothorax occurred probably as a result of mechanical transit of the larvae and inflammatory response in the pleura and the alveoli leading to the formation of sub-pleural blebs.[6] This hypothesis could probably be extrapolated to our case also.

Strongyloidiasis presenting as an acute exacerbation of COPD has also been reported.[7-9] Non-specific signs and symptoms, use of corticosteroids for managing the exacerbations which blunt the eosinophilic response in addition to indiscriminate self-medication with corticosteroids by COPD patients enhance the possibility of hyperinfection syndrome and disseminated stronglyoidiasis and pose a major challenge in diagnosing and managing these patients.

Diagnosis of strongyloidiasis is mainly by the demonstration of eggs and larvae in stool specimens.[3] In patients with hyperinfection syndrome, larvae have been identified in sputum, endotracheal aspirates, bronchoalveolar lavage specimens, pleural fluid and transbronchial and open lung biopsies. Despite lack of specificity, presence of serum precipitins immunoglobulin G (IgG) against *Strongyloides stercoralis* is useful in diagnosis and follow up.[10]

Treatment of strongyloidiasis is challenging as presence of even a single parasite can perpetuate the autoinfection cycle. Ivermectin is the drug of choice in a dose of 200mcg/kg per day until stool examination remains negative for two weeks.[11]

In conclusion, since the clinical and radiological manifestations are varied and non-specific, a high index of suspicion is of prime importance to diagnose strongyloidiasis. In patients residing in or having travelled to endemic areas even in the remote past, with any of the above-mentioned risk factors, presence of eosinophilia with pulmonary infiltrates or patients with gastrointestinal and pulmonary manifestations should prompt suspicion of the disease. The diagnosis is often made by finding of larvae in respiratory specimens or stool examination as highlighted in our case. Early diagnosis and treatment including anti-helminthics and reduction of immunosuppressive drugs where appropriate, may be life-saving in patients with hyperinfection syndrome/disseminated strongyloidiasis, which otherwise can be fatal.

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