Case Reports

Strongyloidiasis—a forerunner of adult T-cell lymphoma

Introduction

Strongyloides stercoralis is an intestinal nematode that affects more than 100 million people worldwide especially in tropical regions such as Southeast Asia, South America and sub-Saharan Africa. Gastrointestinal infestation may vary from asymptomatic to ileus or intestinal obstruction in patients with heavy intestinal infestation. Though Strongyloides infection can occur in immunocompetent adults, a search for an underlying immunosupression may result in early diagnosis and institution of therapy in diseases such as lymphoma[1]. On the other hand, a few studies have shown that Strongyloides hyperinfection may result in the development of adult T-cell leukaemia/lymphoma[2-4]. We report two patients of Strongyloides infection with different modes of gastrointestinal presentation, who later developed T cell lymphoma while on follow-up.

Case report 1

A 56-year-old man presented with chronic, large volume watery diarrhoea of 3 months' duration. There was no blood or mucus in the stools. He had pedal oedema and a weight loss of 4 kg during this period. There was no past history of medical or surgical illness. On clinical examination he had pedal edema. He had no lymphadenopathy or visceromegaly. Laboratory investigations showed a leukocyte count of 11400 cells/mm³ with 8% eosinophils. His total serum protein level was 4.3 g/dL with an albumin level of 1.8 g/dL. Other biochemical investigations were normal and enzyme-linked immunosorbent array (ELISA) test for human immunodeficiency virus (HIV) was negative. His duodenal biopsy showed S. stercoralis (**Figure 1**). He was treated with four doses of 12 mg ivermectin on days 0, 1, 15 and 16; following which he improved symptomatically. Thirteen months later, he presented with generalized lymphadenopathy of one month duration and altered sensorium of one week duration. His clinical examination revealed enlarged cervical, submandibular, axillary and inguinal lymph nodes of sizes varying from 2–3 cm in diameter. He also had pedal oedema and hepatosplenomegaly. Laboratory investigation showed: haemoglobin level of 13.6 g/dL, leukocyte count of 62000 cells/mm³ (63% lymphocytes), erythrocyte sedimentation rate of 48 mm/hour, alkaline phosphatase levels of 784 U/L and serum albumin levels of 3 g/dL. Peripheral smear showed atypical cells with irregularly cleaved nuclei and occasional cells with flower-like nuclei. Lymph node biopsy was consistent with adult T-cell lymphoma (**Figure 2**). The patient's condition deteriorated after 24 hours of admission and he succumbed to cardiorespiratory failure.

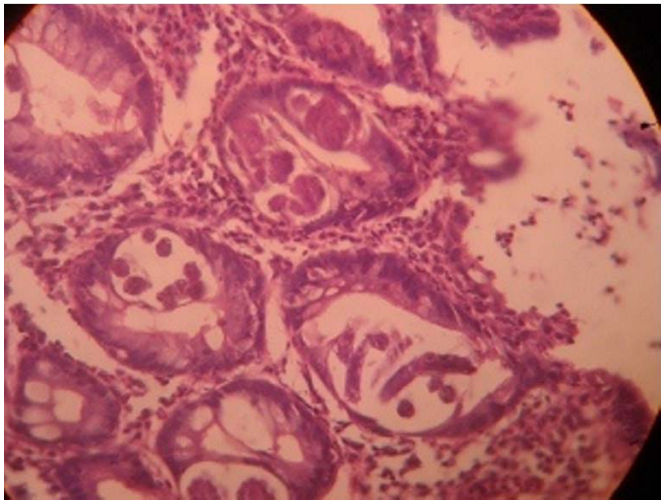

Figure 1: Duodenal biopsy showing S. stercoralis.

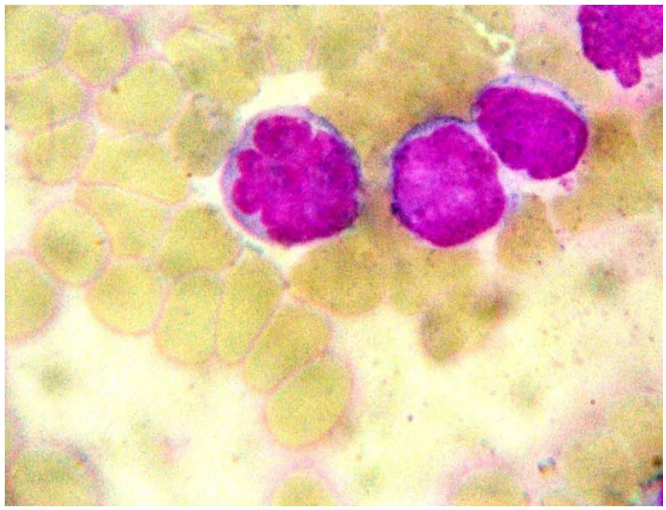

Figure 2: Peripheral smear showing cells with flower-like nuclei in adult T-cell lymphoma.

Case report 2

A 48-year-old woman presented with periumblical, colicky, postprandial abdominal pain of 8 months' duration, with bilious vomiting and constipation of 2 weeks' duration. She had a history of impaired fasting glucose detected recently and was

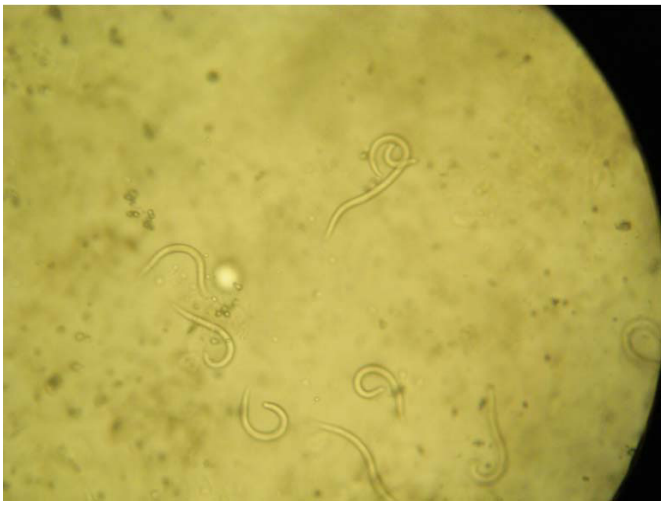

Figure 3: Vomitus containing S. Stercoralis larvae.

advised dietary modification. Her clinical examination revealed a body mass index of 17.2 kg/m² with pallor, glossitis, cheilitis and tinea versicolor. She had a firm hepatomegaly. Laboratory investigations showed peripheral eosinophilia, erythrocyte sedimentation rate of 92 mm/hour, hypochromic microcytic anaemia in peripheral smear, and hypoalbuminaemia (2.7 g/dL). Rest of her laboratory investigations and ELISA test for HIV were negative. Contrast-enhanced computed tomography (CECT) scan showed minimal circumferential, irregular wall thickening in the junction of the second and third part of the duodenum with mild luminal narrowing. Her upper gastrointestinal endoscopy showed granular whitish appearance of the duodenum with narrowing at the periampullary region. Duodenal biopsy showed chronic duodenitis and mild tissue eosinophila with numerous strongyloides larvae. Her stool and gastric aspirate also showed strongyloides larvae (**Figure 3**). Bone marrow examination done to rule out any indolent malignancy was normal. She was treated with 12 mg ivermectin for 2 consecutive days repeated after 2 weeks. She improved with no further episodes of vomiting and was kept on follow-up. Six months later, she presented with abdominal pain and nausea. On examination, she was found to have generalized lymphadenopathy with enlarged cervical, axillary and intra-abdominal lymph nodes. Stool microscopy and duodenal aspirate was negative for strongyloides. Lymph node biopsy revealed adult T-cell lymphoma and she was started on chemotherapy.

Discussion

S. stercoralis is unique among intestinal nematodes in its ability to complete its life cycle within the host through an asexual autoinfectious cycle. The autoinfection cycle is normally prevented by the host immune system, most notably by the Th2 response. S. stercoralis is capable of transforming into a fulminant illness especially when there are defects in the cell-mediated immunity.[1] Steroid therapy and human T-lymphotropic virus-1 human T-lymphotropic virus-1 ((HTLV-1) infection are the most consistent associations. The altered cytokine milieu in cases of lymphoreticular and haematological malignancies may explain how these conditions result in S. stercoralis hyperinfection. Although acquired immunodeficiency syndrome (AIDS) and malnutrition have both been associated with hyperinfection, the number of reported cases is much less than expected.

The association between adult T-cell lymphoma and strongyloidiasis has been reported in the world literature.[2–4] In both the cases described above, the patients presented with T-cell lymphoma after one year of strongyloidiasis. Patients co-infected with HTLV-I and S. stercoralis have higher levels of interferon (IFN)-gamma and lower levels of interleukin (IL-5) and IL-13. This leads to a decreased S. stercoralis-specific type 2 immune response.[5] Clinically, patients coinfected with HTLV-I and S. stercoralis may develop severe strongyloidiasis, as well as a low cure rate in response to anti-S. stercoralis drugs.

On the other hand, some studies have shown that Strongyloides hyperinfection may result in the development of adult T-cell leukaemia/lymphoma. This may be due to a Strongyloides antigen that induces a potent polyclonal T cell mitogenic response, and reactivation of HTLV-1 expression.[6,7] The alternative hypothesis is that Strongyloides hyperinfection causes general immune suppression that permits HTLV-1 replication and spread. Expansion of infected T-cells may enhance genetic instability resulting in inactivation of DNA repair mechanisms, predisposing to cancer.[8,9] On the basis of the above studies and our case series, we recommend a close follow-up of patients with Strongyloides hyperinfection with an initial HTLV-1 serology, and a detailed clinical examination and haemogram every 3 months after the diagnosis.

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Tubercular pancreatic abscess: diagnostic dilemma and management

Introduction

Cystic lesions of the pancreas are most commonly pseudocysts, pancreatic abscesses or cystic neoplasms.[1] Isolated pancreatic tuberculosis is uncommon.[2] Tubercular pancreatic abscess is a rare entity and, therefore, not usually considered in the differential diagnosis of a cystic pancreatic lesion. We report a rare case of isolated tubercular pancreatic abscess diagnosed on the basis of a biopsy of the abscess wall.

Case report

A 50-year-old woman presented with low-grade intermittent fever and upper abdominal discomfort for the past 2 months. She also complained of recurrent episodes of non-bilious vomiting, anorexia and weight loss of 20 kg. She did not give any history of upper abdominal pain during this time. She was malnourished, had mild pallor and generalized oedema. There was no peripheral lymphadenopathy. Examination of the chest did not reveal any abnormality. She had an ill-defined lump in the right hypochondrium and epigastrium. Investigations showed a low haemoglobin level (8 g/dL; reference range: 12–15 g/dL), normal total leukocyte count (6700 cells/mm^3; reference range: 4000–11000 cells/mm^3) and raised erythrocyte sedimentation rate (55 mm in 1st hour; reference range: 0–20 mm in 1st hour). She had a normal serum bilirubin level (0.8 mg/dL; reference range: 0.8–1 mg/dL), raised alkaline phosphatase level (547 IU/L; reference range: 80–240 IU/L) and a low serum albumin level (2.1 g/dL; reference range: 4–5.5 g/dL). Hydatid serology was negative. Chest X-ray was normal. Ultrasound abdomen showed a 13 cm × 7.6 cm heterogeneous collection with thick septae in the region of the head of the pancreas. Contrast-enhanced computed tomography (CECT) scan showed a large, septated cystic lesion replacing the head and uncinate process of the pancreas extending to the porta hepatis (**Figure 1**). The rest of the pancreas and liver were normal (**Figure 2**). The small bowel was normal and the mesenteric and retroperitoneal lymph nodes were not enlarged. There was no dilatation of the biliary or pancreatic ducts. With a working diagnosis of an infected cystic lesion of unknown aetiology, intravenous broad-spectrum antibiotics were started following which her symptoms improved but did not resolve. Ultrasound-guided diagnostic aspiration of the cystic lesion yielded purulent fluid. Stain for acid-fast bacilli (AFB) was negative. The amylase levels in the fluid were higher than the upper limit of the normal serum amylase levels (250 U/L; reference range: 0–95 U/L). Culture of the fluid was sterile for bacteria. Fluid cytology was non-contributory. Cyst fluid carbohydrate antigen (CA) 19–9 (25 U/mL; reference range: 0–37 U/mL) was not raised.