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Hypereosinophilia with eosinophilic colitis as first manifestation of peripheral T cell lymphoma: a case report

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ABSTRACT

Peripheral T cell Lymphoma (PTCL) is a rare variety of non-Hodgkin's Lymphoma with poor prognosis. Besides lymphadenopathy, this malignancy can present with different atypical features, which may baffle clinicians. We here present the case of a 59 year old male who presented with chronic diarrhoea. This was proved to be eosinophilic colitis on colonoscopy. Later, he developed peripheral hypereosinophilia (absolute eosinophil count: 36252/μL) and abdominal lymphadenopathy (mainly retroperitoneal and right iliac). Biopsy and immunophenotyping from the lymph nodes proved this to be a case of PTCL. The patient also had eosinophilic pleural effusion and ascites. He was started on E-CHOP chemotherapy, which led to complete reversal of the peripheral eosinophilia. The eosinophilic colitis responded partially to steroids. Hypereosinophilia has been reported in PTCL but such eosinophilic colitis and eosinophilic pleural effusion are extremely rare.

INTRODUCTION

Peripheral T cell lymphoma (PTCL) is a subtype of non-Hodgkin's lymphoma (NHL) with a distinctive poor prognosis. [1]This haematological malignancy is comparatively rare, with USA registry data showing an incidence of <1 per 100000. [1] There are many subtypes of PTCL and their relative abundance vary geographically. [1]The commonest variety is PTCL-NOS. [1]PTCL usually presents with lymphadenopathy, like other varieties of NHL. [2] But sometimes atypical presentations may be present which can confuse clinicians. [2]We here present a case of PTCL presenting with peripheral hypereosinophilia and involvement of multiple systems with eosinophil infiltrates.

THE CASE REPORT

A 59 year old male first came to the general medicine clinic with complaints of recurrent diarrhoea for three months. He had never had any abdominal complaints in the recent past and was not on any drugs. His initial blood tests were normal; stool tests only revealed the presence of occult blood. Ultrasonography of abdomen showed only thickened caecum. There was no lymphadenopathy or ascites. As a part of the work up, he also underwent a colonoscopy which showed scattered ulcers with wall thickening throughout the colon. Biopsy was also taken from those ulcers which showed (figure 1) inflammatory infiltrates in the mucosa with a predominance of eosinophils. There was no evidence of any parasite or malignancy. The patient was diagnosed as a case of eosinophilic colitis and started on oral steroids.

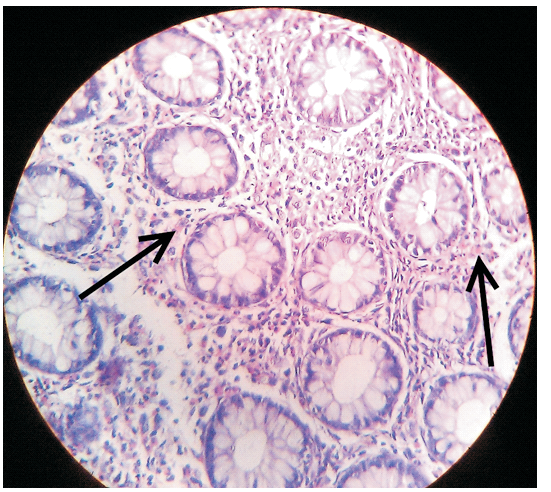

Figure 1 : Colonic mucosa biopsy of the patient showing inflammation along with infiltration by eosinophils (black arrows)

After two months, in May 2017, the patient was again

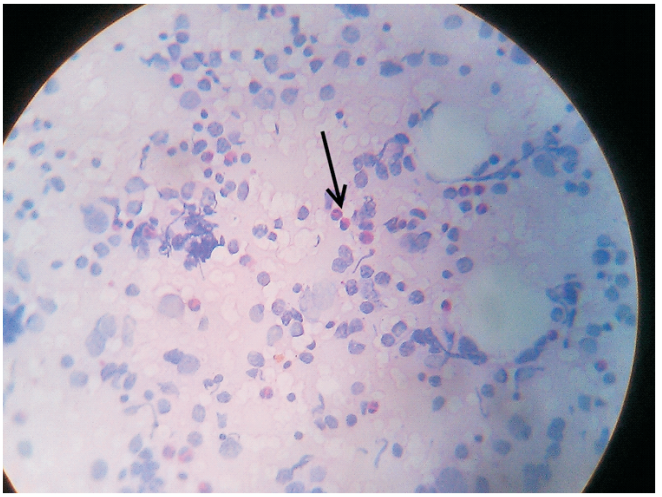

Figure 2 : Ascitic fluid cytology showing plenty of eosinophils (black arrow)

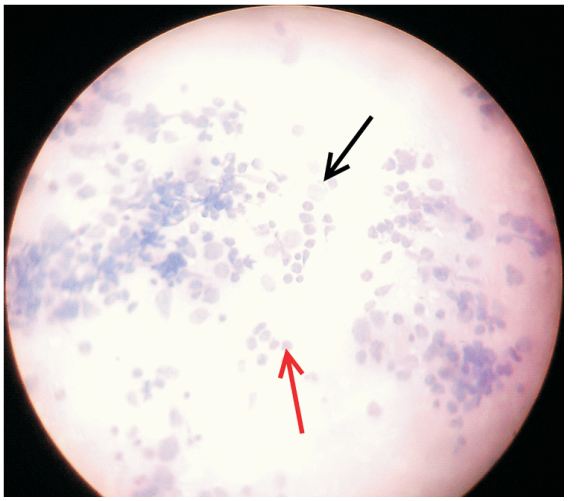

Figure 3 : FNAC slide from the right iliac fossa mass showing large atypical lymphoid cells (black arrow) and eosinophils (red arrow)

admitted in our unit with fever and loss of weight. His bowel symptoms had resolved partly but he presented with loss of about 10 Kg in weight. Clinical examination revealed a BMI of 16.3, mildly elevated temperature, moderate pleural effusion on the left side and an ill-defined firm, tender mass in the right iliac fossa. On questioning, the patient revealed that he had felt a lump in the right lower abdomen for the last one week. Shifting dullness was also present in the abdomen. There was no other lymphadenopathy.

Initial laboratory reports revealed hemoglobin of 12 g/dl, total leukocyte count of 48990/µL and an eosinophil percentage of 74. Absolute eosinophil count was 36252/µL. Platelet count was 158000/µL. Except for the abundant eosinophils, peripheral smear was normal. Blood urea and electrolytes, liver function test and sugar levels were normal. Serum LDH level was 1400 IU/L (N: 120-250). Ultrasonography of abdomen was done which showed presence of ascites, retroperitoneal lymphadenopathy (largest~3 cm in diameter) and an ill-defined mass, separate from the colon, in the right iliac fossa. Also, there was significant pleural effusion on the left side. There was no hepatosplenomegaly. Pleural fluid analysis revealed cell count of 5500/cmm with 70% eosinophils. Pleural fluid protein was 5.6 g/dl, LDH was 858 IU/L and ADA level was 21.9 IU/L. Ascitic fluid study was revealed significant eosinophilia (figure 2). Pleural and ascitic fluid Pap smear for malignant cells was negative.Echocardiography with Doppler did not show any evidence of cardiomyopathy. HIV, hepatitis serology and anti-filarial antibodies were negative. Connective tissue markers, including ANF, ANCA and anti-dsDNA were also negative. A repeat colonoscopy was done which showed persistence of some of the ulcers seen earlier. Contrast enhanced CT scan of thorax and abdomen was done which showed lymphadenopathy only in the areas already seen by USG scan. There was a mass in right iliac fossa, separate from caecum. No thoracic or mediastinal lymph node enlargement was seen. VDRL and Toxoplasma IgG were also negative.

At first, an FNAC was done from the right iliac fossa mass which revealed (figure 3) eosinophils, large atypical mononuclear cells and reactive lymphocytes. Any definite diagnosis could not be reached but the features were suggestive of lymphoproliferative disorder. A CT guided biopsy was done from the same mass again, which showed (figure 4) replacement of normal lymph node architecture with plenty of atypical

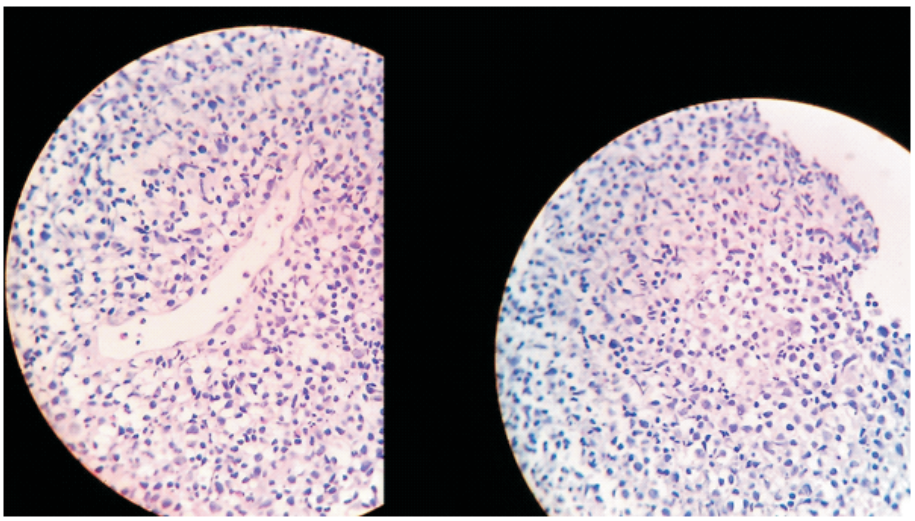

Figure 4 : Abdominal Lymph node biopsy showing effacement of normal nodal architecture with numerous small lymphoid cells with hyperchromatic nuclei and high N/C ratio

lymphocytes with some invasion of blood vessels. The cells were positive for CD3 and 4 and some cells were positive for CD7. Some were dual positive for CD4 and 8. CD15 and 30 were negative. Also, B cell markers, CD19 and 20 were negative. The pathologist opinion was a case of peripheral T-cell lymphoma (PTCL), an aggressive variety of NHL. Thus, finally, the diagnosis was a case of PTCL with hypereosinophilia in peripheral blood, eosinophilic pleural effusion and eosinophilic colitis.

After a multi-disciplinary meeting, the patient was started on the E-CHOP regimen every 21 days. After the first cycle, he had remarkable change in peripheral blood picture with total leukocyte count reducing to 8000/µL and 5% Eosinophils. Also, he had regression of the palpable mass in right iliac fossa. His fever also subsided. But unfortunately, after the 3rd cycle, he had severe pancytopenia and developed a sepsis. He succumbed to the sepsis later.

DISCUSSION

Hypereosinophilia is a syndrome with diverse aetiologies. [3]It can occur as a primary syndrome or may occur secondary to other diseases. Atopic disorders, parasites, drug effects and malignancy have been reported to cause hypereosinophilia. [3] Of these, malignancy is a comparatively rare cause of hypereosinophilia. Haematological malignancies causing hypereosinophilia is even rarer. [3]The increase in eosinophils is considered a paraneoplastic phenomenon (except for eosinophilic leukemia) and reported cases have shown that it generally remits quickly with onset of chemotherapy. [3, 4]In our case too, the absolute eosinophil count decreased by more than 95% with only one dose of E-CHOP therapy.

Lymphomas have been reported rarely to be associated with eosinophilia. The exact mechanism of eosinophilia in various types of lymphoma is unknown but it is hypothesized to be related to alteration in IL-5 pathways. [3, 5]In Hodgkin's Lymphoma (HL), eosinophilia is comparatively more common. [5]Eosinophils are an important element in the pathobiology of HL and increased eosinophil count also has a prognostic significance in HL. [5]In NHL, hypereosinophilia is more common in T-cell variants than the B-cell varieties. [6]

Besides the lymphadenopathy, PTCL can have atypical manifestations. There may be dermatosis, neuropathy, hand swelling and some haematological changes. [2, 7]The haematological changes in PTCL can vary from hypereosinophilia, increased fraction of hypodense neutrophils to neutrophilia and thrombocytosis. [3, 8, 9]

The hypereosinophilia in PTCL may be limited to the peripheral blood or may also involve other organs. [3]Infiltration of other organs by reactive eosinophils may or may not be associated with infiltration with the malignant T cells. In published case reports of PTCL, biopsy of the bone marrow and liver have showninfiltration by eosinophils only without any evidence of malignant cell infiltration. [3]There may be serous cavity effusions too. In one published case report, the pleural fluid analysis demonstrated malignant cells. [3] But in our case, the pleural fluid showed hypereosinophilia only without any malignant cells. The paraneoplastic hyper-eosinophilia in lymphoma may also cause CVA. [10]

The eosinophilic colitis in our case is unique. Other published case reports have shown colonic involvement by the PTCL cells, leading to diarrhoea. [11] But in our case, the diarrhoea was due to eosinophilic infiltration of the colon only. The colonic biopsy did not show any malignant cell infiltration. Thus, in addition to the extreme hypereosinophilia, our case had two other unique features: the eosinophilic colitis and eosinophilic pleural effusion.

CONCLUSION

PTCL may sometimes present with atypical features involving the gastrointestinal system and serous cavity effusions. Even if any evidence of malignancy is not present at the initial stage of an eosinophilia, follow up should be done for subsequent appearance of new clinical features.

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