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Case Report of Chronic Eosinophilic Leukemia Associated with Long-Term Use of Infliximab

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Abstract

Hypereosinophilic syndrome (HES) is a rare condition in which the quantity of eosinophils in the body's blood and tissues increases substantially. Hypereosinophilic syndrome can often go underdiagnosed as the symptoms such as cough, malaise, chills, night sweat, and flu-like symptoms can often be attributed to allergic causes. We present a patient who has been diagnosed with Hypereosinophilic syndrome, later determined to be chronic eosinophilic leukemia, after long-term treatment with infliximab for ulcerative colitis. Follow-up immunochemistry showed that discontinuation of infliximab improved the polyclonality of eosinophils in our patient. Further research and reported cases will help us better understand this relationship.

Keywords: Hypereosinophilic Syndrome; Chronic Eosinophilic Leukemia; Infliximab; *i*matinib; Bone Marrow Biopsy; Diagnosis; Epidemiology; Chemotherapy.

Introduction

Hypereosinophilic syndrome (HES) is a rare disorder in which there is an unidentified increase in the number of eosinophils in the blood and tissues throughout the body. The criteria to determine this syndrome include eosinophilia (≥1500/mm^3) in the blood for greater than six months, no evidence indicating there may be a parasitic infection, allergic reaction, or any other common cause of eosinophilia, and signs that systemic organs may be involved depending on the patients presenting symptoms [1]. Two variants of Hypereosinophilic syndrome have been identified with varying underlying etiology. These subtypes are known as the lymphocytic variant and the myeloproliferative variant of chronic eosinophilic leukemia [2]. Hypereosinophilic syndrome primarily occurs in persons between the ages of 20 and 50, more often in males than females, though cases have been rising in the children and infant population [3]. Though, there is a lack of population-based data, the incidence of Hypereosinophilic syndrome reported by the Surveillance, Epidemiology, and End Results (SEER) database is known to range from 0.3 to 6.3 per 100,000 [4]. Clinical manifestations of the syndrome present similarly in all ages, with the most common presenting symptoms including rash, myalgias, fatigue, cough, dyspnea, and retinal lesions [5]. We present a patient diagnosed with Hypereosinophilic syndrome, later determined to be chronic eosinophilic leukemia, after long-term treatment with infliximab for ulcerative colitis.

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Case Presentation

A 44-year-old male presents to the office with a past medical history of ulcerative colitis for seven years. The patient was actively treated with infliximab and mercaptopurine for the past seven years. The patient presented with a cough, malaise, chills, night sweat, and flu-like symptoms. Initially, the patient was treated for possible allergic bronchitis as he has a history of allergies without any improvement. On lab results, complete blood count was increased. White blood cell count was increased to 30,000 with predominant eosinophilia. Computerized Tomography (CT) scan of the chest with contrast showed bilateral apical infiltrates with no adenopathy or mass. Computerized Tomography scan of abdomen and pelvis did not show any hepatosplenomegaly. Bone marrow biopsy showed hypercellular bone marrow with trilineage hematopoiesis and markedly increased eosinophils. The blast cells were less than 1 percent. Surprisingly, a peripheral molecular panel of myeloproliferative disease showed positive platelet-derived growth factor receptor A (PDGFRA), and myeloproliferative leukemia mutation (MPL) was positive, suggesting a diagnosis of chronic eosinophilic leukemia. The patient was receiving infliximab every two months for several years and was suspected as the cause of leukemia. After stopping infliximab and initiating the treatment with imatinib, the patient's condition improved significantly. A follow up immunohistochemistry was performed and showed polyclonal plasma cells rather than monoclonal. This confirmed our suspicion that the chronic eosinophilic leukemia in our patient was associated with chronic use of infliximab.

Discussion

According to the World Health Organization (WHO), the clinical course of Hypereosinophilic syndrome may affect various organs, with dermatologic involvement reported being the highest (69%), followed by pulmonary (44%), gastrointestinal (38%), and cardiac-related issues (20%) respectively [6]. Eosinophils are known to develop from CD34+ hematopoietic stem cells along the myeloid lineage. The cytokine network typically controls the eosinophil levels throughout the body's tissues and organs. When there is an imbalance, T-cells, mast cells, and stromal cells are triggered to release cytokines such as Interleukin-5, Interleukin-3, and granulocyte-macrophage-colony-stimulating factor, and platelet-derived growth factor (PDGF), resulting in exponential growth and activation of eosinophils. This results in a vicious cycle as the activated eosinophils release their own mediators stored in granules to promote growth, negatively influencing homeostasis. The active molecules can damage the surrounding environment leading to fibrosis and thrombosis in various organs [7]. Since there is a likelihood that hypereosinophilic syndrome can affect many organ systems, the diagnostic workup should be thorough and catered to the presenting symptoms of the patient. After taking a careful history and physical exam, a complete blood count with differentials, routine chemistries, electrocardiogram, echocardiogram, pulmonary function tests, chest X-ray, ultrasound or computed tomography, bone marrow aspirate, and tissue biopsies may be ordered. Given the other known causes of an increased number of eosinophils, tests can also be ordered to rule out infection by blood culture, stool culture, serological studies (for human immunodeficiency virus, cytomegalovirus, Epstein barre virus, hepatitis B virus, hepatitis C virus), and imaging studies if necessary [8]. For acute intervention in severe cases, it is essential to treat with a single stat dose of a high-dose glucocorticoid such as prednisone or methylprednisolone once the possibility of a parasitic infection has been ruled out. If unable to rule out a parasitic infection, ivermectin should be administered along with the steroid to prevent possible strongyloides hyper-infection syndrome. After two days, if no sufficient improvement is seen in the hypereosinophilic syndrome or clinical symptoms, second-line treatment should be added to the patient's regimen, such as imatinib for the myeloproliferative variant, until their absolute eosinophil counts have improved. Studies have shown that imatinib, a tyrosine kinase receptor inhibitor, has high efficacy for hypereosinophilic syndrome and other related chronic myeloid disorders [9]. Though asymptomatic patients do not require treatment, they must be closely monitored to avoid organ damage [10].

Conclusion

We report a severely underreported association between infliximab and monoclonal proliferation, leading to, in our case, chronic eosinophilic leukemia. We recommend clinicians remain vigilant regarding symptoms of hypereosinophilic syndrome in long-term users of infliximab. Depending on lab results, additional immunochemistry workup may be necessary to determine monoclonality if leukemia is suspected. Following discontinuation of infliximab and appropriate treatment initiation, an increase in polyclonality may substantiate the suspicion of leukemia secondary to infliximab. We hope that further research and cases are reported in the literature to understand this relationship better.

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Availability of data and material: The datasets generated and/or analyzed during the current study are not publicly available due to HIPAA but are available from the corresponding author on reasonable request, if permissible by patient.

Authors' contributions: 'WM' was a major contributor in the writing of the manuscript. 'HN' oversaw the patient with the attending physician and contributed to the editing of the paper. 'A.A.C' helped with the interpretation of the patient data. All authors approved the final manuscript.

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