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Case Report

Eosinophilic Perimyositis In a Young Male: A Case Report and Review of Literature

Bimal Patel

Department of Pathology, Gujarat Adani Institute of Medical Sciences, Bhuj, Kachchh, Gujarat

* Correspondence: Dr. Bimal Patel (bimaldoctor@gmail.com)

ABSTRACT

Eosinophilic myositis is rare group of disorder characterized by eosinophilic infiltration of skeletal muscles. It generally presents with pain, swelling and weakness of the affected muscle group or limb. The main cause of skeletal muscle eosinophil infiltration is parasitic infections. Other conditions such as hypereosinophilic syndrome, muscle dystrophies, drugs or toxins can also cause such a condition. However, most cases have no known etiological factor and are considered as idiopathic. Idiopathic eosinophilic myositis can be classified into three subtypes: focal eosinophilic myositis, eosinophilic polymyositis, and eosinophilic perimyositis which can be differentiated by pathological studies. The present report is a case of eosinophilic perimyositis who presented with pain and swelling in the region of right thigh.

Keywords: Eosinophilic Myositis, Eosinophilic Perimyositis, Muscle, Inflammation

INTRODUCTION

The term eosinophilic perimyositis (EP) was coined by Serratrice et al[1] in 1980. They described two patients with a benign relapsing myalgia which involved the legs, with perimysial eosinophilic infiltrates, peripheral eosinophilia, normal creatine phosphokinase levels, and no evidence of systemic involvement. EP is a benign condition that is generally associated with myalgias and mild muscle weakness. EP is a histologically and pathologically diverse group of disorder affecting the muscle or it's supporting connective tissue structures with infiltration of eosinophils.[2]

CASE HISTORY

A 26-year male presented with complaints of pain on the flexural aspect of the right thigh for the past 6 months. Pain used to increase at night and decrease in the morning. Patient used to walk with a limp. Pain used to decrease with NSAID's. There was no associated swelling or redness over the affected skin. For the past 1 month patient developed increased intensity of pain which was associated with swelling over the posterior aspect of thigh. There was no history of weakness of any part of body, numbness, parasthesias or inability to identify touch. There was no history of fever, weight loss, anorexia. There was no history of drug intake or exposure to toxins, dye, organic or inorganic substances. There was no history of a visit to sea shore area in near future. The patient was a vegetarian. There was no other significant history.

On examination, his vitals were normal and he was afebrile. There was a localized swelling of 10 x 5 cm over posterior aspect of lower thigh. There was erythema over the involved skin and it was warm and tender to touch. Rest of the general physical and systemic examination including nervous system was normal.

Investigations revealed normal complete blood count with total leucocyte count of 6.7 x 103/cumm with differential count showing neutrophils 67%, lymphocytes 20%, eosinophils count of 3% and monocytes 10%. Absolute eosinophil count was 201/cumm. Liver and kidney function tests along with urine examination was normal. Serum levels of creatine kinase, aldolase and lactate dehydrogenase enzymes were also normal. MRI of the involved area was done which showed diffuse ill-defined high-signal involving muscles of the posterior compartment of distal thigh and upper leg. There was diffuse band like high signal along the deep fascia predominately at the posterior, posterolateral and medial aspect and extending anteriorly along the posteromedial surface of vastus medialis muscle.

There was similar high-signal intensity and minimum fluid in the intermuscular plane. A biopsy from muscle and surrounding fascia from the affected site was done under general anaesthesia. Peroperatively the muscles were healthy with mild fluid in the posterior compartment. Histopathological examination showed moderately dense perivascular and transmural infiltrates of lymphocytes, eosinophils, histiocytes, few neutrophils and plasma cells on the fascia of the posterior compartment of the right thigh. There was no evidence of necrosis, myophagocytosis, regenerative activity or endomysial inflammation.

There was no evidence of parasite or granulation tissue. Special stains for microorganisms including fungus were negative. In view of eosinophilic infiltration, serum IgE levels were done which were normal. Neurocysticercosis serology was negative. Stool examination for parasites was negative. Anti Neutrophil Cytoplasmic Antibodies levels were normal. Considering the above situation, a diagnosis of eosinophilic perimyositis was made. Patient was started on steroids at a dose of 0.5 mg/kg body weight after which pain and swelling decreased. The dose of steroids was tapered. After 6 weeks of follow up, the patient was pain free with no swelling at the local site.

DISCUSSION

Eosinophilic myositis includes, clinically and pathologically heterogeneous disorders characterized by eosinophilic infiltration of skeletal muscles. However, in the absence of eosinophil muscle infiltration, the damage may be caused due to cytotoxicity caused by major basic protein (MBP) released by eosinophils.[2,3,4]

Causes of eosinophilic myositis include parasitic infections, systemic disorders with hypereosinophilia such as Eosinophilic Granulomatosis with Polyangitis(EGPA), intake of various drugs or toxins and rarely Idiopathic hypereosinophilic syndromes. If there is no identifiable cause then diagnosis of idiopathic inflammatory muscle disorders is made[5]. EM can be classified into three subtypes: focal EM, eosinophilic polymyositis, and eosinophilic perimyositis. These subtypes can be differentiated on the basis of pathological studies.[5]

Trueb et al[6], found the mean age of 43 years among patients with EP. In this study group, it was more commonly seen in males as compared to females with a male to female ratio of 3.3:1. Myalgias and mild muscle weakness are usually the symptoms seen in patients with perimyositis. It is generally a benign condition. Histopathological examination in such patients shows that muscle fibers are normal and there is perimysial eosinophilic infiltrate at the muscle surface that extends into the fascia[5]. Peripheral blood eosinophilia may be variable.

Eosinophilic perimyositis is diagnosed based on the proposed criteria for diagnosis of patients with suspected eosinophilic myositis[5] as below -

Eosinophilic perimyositis-

Major
1. Myalgias, proximal mild weakness
2. Eosinophilic infiltrate confined to fascia and superficial perimysium, absence of myofiber necrosis
Minor
a) Absence of systemic manifestations
b) Normal creatine kinase and aldolase levels
c) Eosinophilia > 0.5 X 109/l
Exclusion
(i) Toxic oil syndrome, myalgia-eosinophilia, exposure to inorganic or organic substances
**The presence of both major criteria or major criteria number 2 and two minor criteria satisfies the diagnostic criteria.

Corticosteroid therapy may be needed to improve symptoms.[5]

Very few cases of eosinophilic perimyositis have been described so far. Dunand et al[7] described a case of EP as the presenting feature of a monoclonal T-cell expansion. Zivkovic et al[8] described a case of chronic eosinophilic perimyositis with persistent myalgias. Trueb et al[6] described cutaneous manifestations in 13 patients with EP. Serratrice et al had published a case of localized EP[9] and relapsing EP.[10] The present patient fulfilled both major and 2 out of 3 minor criteria for the diagnostic criteria for EP. Other causes of eosinophic myositis/perimyositis were excluded. Patient was started on steroid therapy after which, his symptoms improved dramatically.

CONCLUSIONS

Eosinophilic perimyositis is a rare disease, the diagnosis of which is mainly pathological. It is generally a benign condition which may be treated with steroids.

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