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UNMASKING THE TIGER MAN: WHEN EOSINOPHILIC MYOSITIS MIMICS SARCOIDOSIS

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ABSTRACT

Introduction: Idiopathic inflammatory myopathies are a heterogeneous group of disorders with overlapping clinical and imaging features. Advanced imaging such as ^{18}F-fluorodeoxyglucose positron emission tomography-computed tomography (^{18}F-FDG PET-CT) is increasingly used. The "tiger man" sign is a rare nuclear imaging sign associated with sarcoid myopathy and has been previously described in only two case reports of non-sarcoid myopathy. Eosinophilic fasciitis and myositis lie within a spectrum of eosinophil-associated disorders, for which evidence-based treatment remains limited. In this context, histopathological insights may offer significant diagnostic and therapeutic value.

Case description: A 68-year-old male presented with pain and oedema of the lower legs and forearms, accompanied by recurrent nocturnal fever. He had a prior diagnosis of eosinophilic fasciitis two years earlier. Proximal muscle weakness of the upper legs was noted. Laboratory tests revealed systemic inflammation and elevated creatine kinase (CK) levels, suggestive of an inflammatory myopathy, while screening for infections and autoimmune serology was negative. 18F-fluorodeoxyglucose positron emission tomography-computed tomography (^{18}F-FDG PET-CT) imaging demonstrated a "tiger man" sign, characterized by extensive patchy ^{18}F-FDG uptake in the skeletal muscles of all four extremities. Histopathological analysis confirmed a T-cell-mediated, necrotizing eosinophilic myositis, with no evidence of sarcoidosis or malignancy. Treatment with tacrolimus and plasma exchange resulted in significant clinical improvement and regression of ^{18}F-FDG PET abnormalities.

Conclusion: This is the third report of the "tiger man" sign in a patient with a non-sarcoid myopathy, thereby expanding the differential diagnosis for this imaging finding. Whole-body imaging can identify hypermetabolic yet clinically silent muscular regions, thereby guiding a more accessible and relevant site for biopsy. Histopathological findings can enhance diagnostic certainty and guide a more effective treatment.

KEYWORDS

Eosinophilic myositis, tiger man sign, plasma exchange, tacrolimus

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LEARNING POINTS

- A "tiger man" sign on ^{18}F-fluorodeoxyglucose positron emission tomography-computed tomography imaging is a rare and characteristic sign in sarcoid myopathy, but it is not disease-specific.
- Whole-body imaging can identify hypermetabolic yet clinically silent muscular regions, thereby guiding a more accessible site for biopsy.
- Histopathological analysis can enhance diagnostic certainty and guide a more effective treatment in the context of difficult to treat or idiopathic myositis.

INTRODUCTION

Idiopathic inflammatory myopathies represent a heterogeneous group of disorders with overlapping clinical and imaging features, often complicating accurate and timely diagnosis. In this context, advanced imaging modalities such as ^{18}F-fluorodeoxyglucose positron emission tomography-computed tomography (^{18}F-FDG PET-CT) are increasingly used. Eosinophilic fasciitis and myositis are considered part of a spectrum of eosinophil-associated disorders with overlapping features, and current evidence guiding their treatment remains limited. Histopathological insights may offer significant diagnostic and therapeutic value.

CASE DESCRIPTION

A 68-year-old male was admitted due to pain and edema of the lower legs and forearms in combination with recurrent nightly spiking fever up to 40°C. Two years prior he was diagnosed with eosinophilic fasciitis, based on skin thickening of the forearms and lower legs, peripheral eosinophilia (1300/µl, reference range 30-350/µl) and compatible pathology findings. He was treated successfully with methotrexate (20mg SC qw) and short-term corticosteroids.

At presentation, we noted no relevant skin findings and mild muscle weakness (MMT8 score 144/150, affecting Mm quadriceps and Mm glutei maximi bilaterally). The patient developed progressive weight loss (8% of body weight). Laboratory analysis revealed raised inflammatory markers (C-reactive protein 91.6 mg/l, reference range <5 mg/l, erythrocyte sedimentation rate 71 mm/h, reference range 1-10 mm/h, ferritin 3361 µg/l, reference range 30 - 400 µg/l) and elevated creatinine kinase (CK) levels (1425 U/l, reference range <190 U/l).

^{18}F-FDG PET-CT showed multiple FDG-avid lymph nodes and extensive, superficial and patchy hypermetabolic foci in the skeletal muscles of all four extremities *(Fig. 1)*, known as "tiger man" appearance/sign, reportedly associated with sarcoid myopathy. This characteristic PET-CT image with diffuse (linear and/or patchy) FDG - uptake along the skeletal muscle bundles, thereby mimicking the coat of a tiger, was first described in 2012 by Wieërs et al.[1] and confirmed in multiple subsequent case reports (full list available in appendix).

Repeat muscle-fascia biopsy showed locally extensive and deep T-lymphocytic infiltrates, extending into the endomysium, in the presence of some eosinophilic granulocytes and signs of muscle fiber necrosis, compatible with immune-mediated necrotizing, eosinophilic myositis. No fascial inflammation was observed. As no granulomas were detected and thoracic imaging did not show hilar or mediastinal adenopathy, sarcoidosis was ruled out. Treatment was initiated with plasma exchange (PLEX), (five PLEX sessions every other day) in combination with tacrolimus (target level 8-10 µg/l), leading to a fast clinical response and clear regression of ^{18}F-FDG PET-CT abnormalities *(Fig. 1)*. The decision to initiate PLEX was driven by the severity of the clinical presentation and the possibility of a concomitant infection. No relevant adverse events were noted during follow-up.

Eosinophilic fasciitis and eosinophilic myositis are considered part of a spectrum of eosinophil-associated

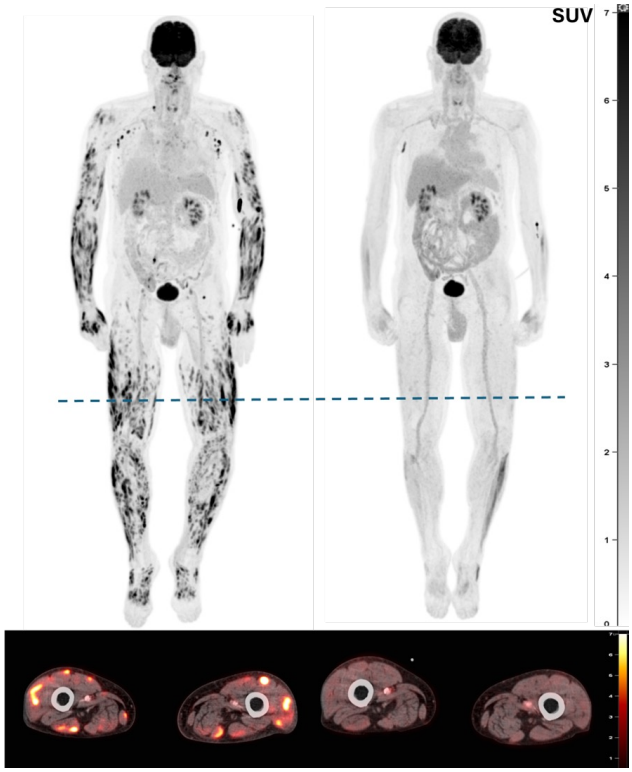


Figure 1. *^{18}F-fluorodeoxyglucose positron emission tomography-computed tomography images at diagnosis (left) and three weeks after initiation of plasma exchange and tacrolimus (right). Lower panel: axial fused positron emission tomography/computed tomography images at the level indicated by the blue dotted line.*

disorders with overlapping clinical, laboratory, and histopathological features. In our patient, the clinical presentation and biopsy findings were consistent with diffuse eosinophilic myositis, one of the three recognized subtypes[2-4]. This subtype is characterized by proximal muscle weakness, elevated CK levels, and deep, extensive eosinophilic infiltration associated with myonecrosis. In contrast, focal eosinophilic myositis is a benign, localized condition involving a single muscle without systemic manifestations, whereas eosinophilic perimyositis presents with myalgia and eosinophilic infiltration stays limited to the fascia and superficial perimysium, without evidence of necrosis.

DISCUSSION

Although a "tiger man" sign on ^{18}F-FDG PET-CT imaging is regarded as characteristic of sarcoid myopathy, it remains a rare finding with less than 20 cases reported in the literature to date. To the best of our knowledge, the present case represents the third report of a "tiger man" sign in a patient with non-sarcoid myositis, highlighting that this distinctive FDG uptake pattern is not disease specific. To date, the only non-sarcoid cases were reported in a patient with dermatomyositis associated with thymoma[5] and in a patient with Sjögren's disease[6]. In the latter case, the "tiger man" sign was less prominent and predominantly observed in the bilateral biceps and pelvic girdle musculature.

The extent of muscle involvement on the ^{18}F-FDG PET-CT was striking, with FDG uptake observed in regions without corresponding pain or oedema. A similar case has been reported in which ^{18}F-FDG PET-CT likewise demonstrated FDG uptake in clinically asymptomatic muscle areas[7]. That patient presented with lower limb swelling and intermittent fever accompanied by elevated creatine kinase levels, and muscle biopsy confirmed eosinophilic myositis. However, in contrast to our findings, the ^{18}F-FDG PET-CT showed focal areas of muscular hypermetabolism rather than a diffuse "tiger man" pattern. The reason why certain hypermetabolic muscle regions remain clinically silent are currently unclear; nevertheless, these observations underscore the importance of performing whole-body ^{18}F-FDG PET-CT imaging.

CONCLUSION

In this case report we present the third case of a non-sarcoid myositis associated with a "tiger man" sign on ^{18}F-FDG PET-CT, thereby expanding the differential diagnosis of this rare sign beyond sarcoidosis. Whole-body imaging can identify hypermetabolic yet clinically silent muscular regions, thereby guiding a more accessible and relevant site for biopsy. The relevance of histopathological findings is highlighted, as these can enhance diagnostic certainty and guide a more effective and tailored treatment.

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APPENDIX

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