Clinical note

\(^{18}\text{F}-\text{FDG PET/CT}\) in diagnosis and response evaluation in an unusual case of antisynthetase syndrome presenting as pyrexia of unknown origin

T.K. Jain\(^{a,}\), R.K. Basher\(^{a,}\), A. Bhattacharya\(^{a}\), B.R. Mittal\(^{a}\), J. Shukla\(^{a}\), M. Prakash\(^{b}\)

\(^{a}\) Department of Nuclear Medicine, PGIMER, Chandigarh 160012, India
\(^{b}\) Department of Radiodiagnosis, PGIMER, Chandigarh 160012, India

A R T I C L E   I N F O

Article history:
Received 15 July 2015
Accepted 12 August 2015
Available online 10 December 2015

Keywords:
Anti-synthetase syndrome (ASS)
Anti Jo-1 antibody
Polymyositis
FDG PET/CT

A B S T R A C T

Anti-histidyl (Jo-1) antibodies have been implicated in the pathogenesis of anti-synthetase syndrome (ASS). A case is presented of a 55-year-old male patient presenting with pyrexia of unknown origin and inconclusive routine investigations. \(^{18}\text{F}-\text{FDG PET/CT}\) was performed to locate any abnormal focus, which showed increased FDG uptake in the proximal shoulder muscles, as well as lung lesions. Subsequent investigation showed the presence of anti Jo-1 antibody, and diagnosed as an anti-synthetase syndrome. The patient was successfully treated with glucocorticoids and cyclophosphamide, and the response was assessed with symptomatic relief and disappearance of FDG uptake in lung and muscle lesions on post-treatment FDG PET/CT.

© 2016 Elsevier España, S.L.U. and SEMNIM. All rights reserved.

18F-FDG PET/TC en el diagnóstico y evaluación de la respuesta en un caso inusual de antisintetasa síndrome que se presenta como fiebre de origen desconocido

R E S U M E N

Los anticuerpos antihistidil (Jo-1) están implicados en la patogenia del síndrome antisintetasa. Presentamos un varón de 55 años con fiebre de origen desconocido y exploraciones de rutina no concluyentes. La \(^{18}\text{F}-\text{FDG PET/TC}\), realizada para la localización de un foco patológico mostró, un aumento de la captación de FDG en la musculatura proximal de los hombros y en lesiones pulmonares. La investigación posterior demostró la presencia de anticuerpos anti-Jo-1 y se diagnosticó un síndrome antisintetasa. El paciente fue satisfactoriamente tratado con glucocorticoides y ciclofosfamida. La respuesta se evaluó por el alivio sintomático y por la desaparición de la captación de FDG en las lesiones pulmonares y musculares demostrada en la \(^{18}\text{F}-\text{FDG PET/TC}\) post tratamiento.

© 2016 Elsevier España, S.L.U. y SEMNIM. Todos los derechos reservados.

Introduction

Inflammatory myositis associated with interstitial lung disease (ILD) and the presence of anti-synthetase auto-antibodies are the characteristics of anti-synthetase syndrome.\(^1\) Interstitial pneumonitis is the most common manifestation of ASS and it may progress to fibrosis, governing the prognosis of disease and increased mortality rate. Fever (80%) is the most common constitutional symptom followed by asthenia and weight loss.\(^2\) Treatment of antisynthetase syndrome is glucocorticoids which seem to improve the articular, muscular and constitutional symptoms, but few of pulmonary disease seem to be corticosteroid resistant. We report a case of 55 years old man who presented with pyrexia of unknown origin and diagnosed as an antisynthetase syndrome with anti-Jo-1 antibodies and successfully treated with cyclophosphamide and glucocorticoids (Fig. 1).

Case report

A 55-year-old male patient, who was incidentally diagnosed to have Hepatitis B admitted with complaints of fever and myalgia since last 3 months and had a history of previous treatment for fever and dyspnea. Computed tomography of the chest revealed consolidatory changes in both the lung fields and pleural effusion. A diagnostic tap was done which was negative for ADA and patient were discharged on antibiotics. He showed no improvement to antibiotic treatment and re-admitted to investigate the cause of fever. On physical examination, he had a fever with normal pulse and blood pressure. He had mild synovitis of small joints of hands. The skin was normal and there was no evidence of Reynaud’s phenomenon. The rest of the general physical and systemic examinations were inconclusive.

* Corresponding author.
E-mail address: drrajender2010@gmail.com (R.K. Basher).

http://dx.doi.org/10.1016/j.remn.2015.08.013
2253-654X/© 2016 Elsevier España, S.L.U. and SEMNIM. All rights reserved.
At admission, his laboratory tests showed increased erythrocyte sedimentation rate (46 mm/h), CRP-4.8 mg/dl (<0.6), leukocytosis (14,300 cells/cm³) and raised liver enzymes (AST/GPT-152.3/92.8). Peripheral blood smears and serology were negative for the malaria parasite. IgG/IgM antibodies for brucella were negative and repeated blood culture also showed no micro-organism growth. Similarly routine urine examination, urine culture, stool examination for ova/cyst and Clostridium difficile toxin were negative. His HBV viral load was not increased (<3.8 IU/ml) and fungal and HIV serology was also negative. Bronchoalveolar lavage for acid-fast bacilli (AFB) and RK -39 antibodies for kala-azar was also negative. RA factor titer was raised to 1:2 (significant >1:1.16) while ANA/ANCA, LKM and PCA titers were negative and serum ACE levels were mildly raised (84 U/L; normal 8–65).

Ultrasonography (USG) of the abdomen revealed mild hepatomegaly and fatty liver infiltration while compression USG of lower limbs was negative for deep venous thrombosis. Echocardiography revealed mild pericardial effusion with no evidence of endocardial vegetation. A trans-bronchial biopsy showed interstitial fibrosis.

With all these inconclusive investigations for fever of unknown origin whole body 18F-FDG PET/CT was performed to localize any abnormally increased FDG uptake, which revealed faintly FDG avid ground glass opacities and fibrotic changes in bibasilar and subpleural region the lung fields (features suggestive of interstitial lung disease), non-FDG avid pericardial effusion, multiple ill-defined hypodense lesions in the multiple skeletal muscles (mainly shoulder muscles) with faint FDG uptake in the periphery of the lesions. On the basis of FDG PET/CT findings patient was further investigated for myopathy as a cause of fever. Clinical examination showed no evidence of proximal, neck and trunk muscles weakness. Electromyography revealed evidence of myopathy. Autoimmune serology was negative for antibodies against nuclear antigen U1-SNRNP. However, anti-Jo antibodies were positive. Serum ferritin (2209; normal 30–400), CPKtotal (3558 U/L at 370; normal 39–308) and CPKMM (2585 U/L; normal 10–165) were grossly elevated.

In the presence of constitutional symptoms of fever and muscular pain, FDG uptake in the lung lesions (ILD), muscular lesions and pericardial effusion with persistently increased AST/PT levels...
دانلود مقاله

http://daneshyari.com/article/4249671