Incidentally Detected Subclinical Limbic Encephalitis on ^{18}F-Fluorodeoxyglucose Positron Emission Tomography/Computed Tomography in a Patient with Myasthenia Gravis and Sarcoidosis: A Rare Immunological Triad?

Abstract

Limbic encephalitis is the inflammation of the medial temporal lobe structures due to direct infectious or indirect autoimmune etiology. Sarcoidosis is a rare cause of limbic encephalitis, has been reported by some authors. Association has also been reported between sarcoidosis, myasthenia gravis, and limbic encephalitis. We report an interesting case of ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography demonstrating limbic encephalitis and sarcoidosis in a known case of myasthenia gravis.

Keywords: *^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography, limbic encephalitis, myasthenia gravis, sarcoidosis*

Tanigassalam Sindhu, Anwin Joseph Kavanal, Rajender Kumar, Harmandeep Singh, Aman Sharma[1]

Departments of Nuclear Medicine and [1]Internal Medicine, Postgraduate Institute of Medical Education and Research, Chandigarh, India

A 62-year-old-male on treatment for myasthenia gravis with azathioprine and pyridostigmine for 3 years presented with low-grade fever, evening rise of temperature, mild headache, irritability, loss of appetite, and loss of weight for 3 months' duration. The laboratory investigation revealed hypercalcemia (13.5 mg/dl) and an elevated C-reactive protein level of 31.6 mg/dl. His X-ray chest was within normal limits. However, ultrasonography and computed tomography of the abdomen were suggestive of hepatosplenomegaly. His blood and urine cultures were sterile, and his bone marrow was normocellular. Tumor markers were within normal limits Figure 1. The ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography (^{18}F-FDG PET/CT) scan maximum intensity projection (MIP) image (a) showed diffuse hypermetabolism in the enlarged liver (17.3 cm) and spleen (15.4 cm) (solid arrows). The fused PET/CT, corresponding PET, and CT images of the brain (b-d, f-h) showed hypermetabolism in the bilateral mesial temporal lobes (arrowheads), basal ganglia, and thalami suggestive of autoimmune limbic encephalitis. The coronal fused PET/CT and CT images of bilateral lungs (e, i) showed FDG avid ground glass haziness. The fused PET/CT, corresponding PET, and CT images of the pelvic region (j-l) showed FDG avid (SUV max 13.2) lytic lesion with soft tissue component in the left ischial tuberosity (dashed arrows). Biopsy from the left ischial lesion revealed epithelioid granulomatous inflammation with multinucleated Langerhans giant cells and foreign body type giant cells suggestive of sarcoidosis. AFB stain was negative. Furthermore, there is no scan evidence of thymoma which is showed in the axial fused PET/CT image (m).

Based on PET/CT findings and histopathological examination demonstrating encephalitis and sarcoidosis, he was started on steroid therapy, following which fever subsided, appetite improved, and he started gaining weight. The steroid dose was eventually tapered from 60 mg to 5 mg once a day Figure 2. Follow-up ^{18}F-FDG PET/CT MIP image (a) showed normalization of hepatosplenomegaly (solid arrows), axial fused PET/CT, corresponding PET and CT images of the brain (b-d, f-h) showed complete resolution of hypermetabolism involving mesial temporal lobes (arrowheads) and reduction in the

How to cite this article: Sindhu T, Kavanal AJ, Kumar R, Singh H, Sharma A. Incidentally detected subclinical limbic encephalitis on ^{18}F-Fluorodeoxyglucose positron emission tomography/computed tomography in a patient with myasthenia gravis and sarcoidosis: A rare immunological triad?. Indian J Nucl Med 2022;37:199-201.

Address for correspondence:
Dr. Rajender Kumar, Associate Professor, Department of Nuclear Medicine, Postgraduate Institute of Medical Education and Research, Chandigarh - 160 012, India.
E-mail:drrajender2010@gmail.com

Received: 01-10-2021
Revised: 06-12-2021
Accepted: 13-12-2021
Published: 08-07-2022

Access this article online

Website: www.ijnm.in

DOI: 10.4103/ijnm.ijnm_154_21

Quick Response Code:

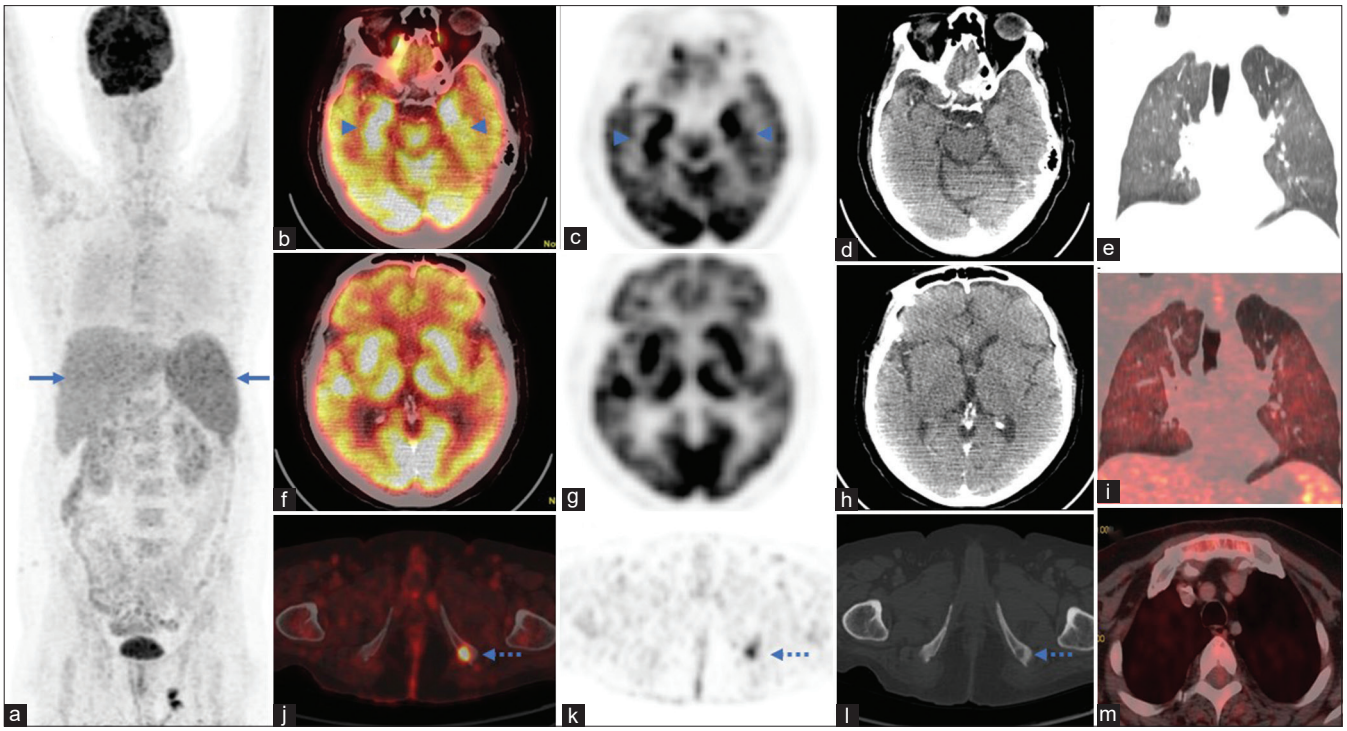

Figure 1: The ^{18}F-FDG PET/CT scan maximum intensity projection (MIP) image (a) showed diffuse hypermetabolism in the enlarged liver (17.3 cm) and spleen (15.4 cm) (solid arrows). The fused PET/CT, corresponding PET, and CT images of the brain (b-d, f-h) showed hypermetabolism in the bilateral mesial temporal lobes (arrowheads), basal ganglia, and thalami suggestive of autoimmune limbic encephalitis. The coronal fused PET/CT and CT images of bilateral lungs (e,i) showed FDG avid ground glass haziness. The fused PET/CT, corresponding PET and CT images of the pelvic region (j-l) showed FDG avid (SUV max 13.2) lytic lesion with soft tissue component in the left ischial tuberosity (dashed arrows). Biopsy from the left ischial lesion revealed epithelioid granulomatous inflammation with multinucleated Langerhans giant cells and foreign body type giant cells suggestive of sarcoidosis. AFB stain was negative. Furthermore, there is no scan evidence of thymoma which is showed in the axial fused PET/CT image (m).

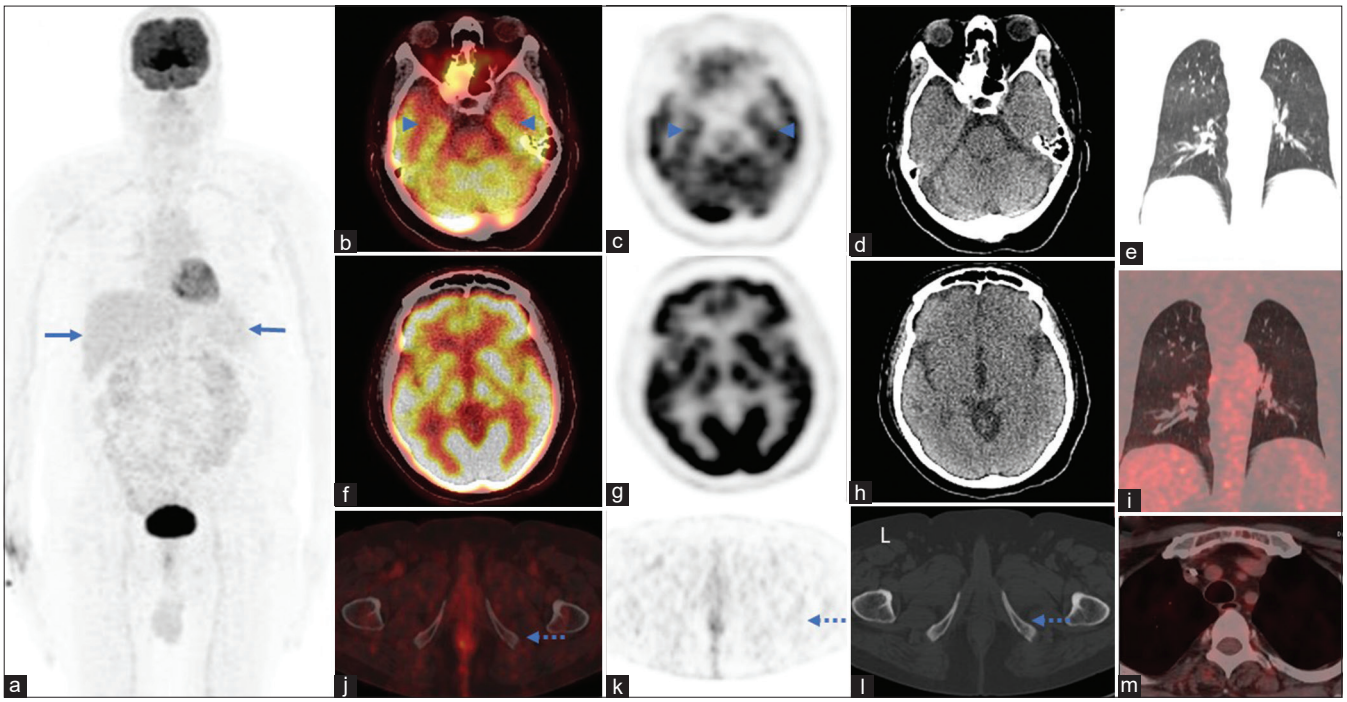

Figure 2: Follow-up ^{18}F-FDG PET/CT MIP image (a) showed normalization of hepatosplenomegaly (solid arrows), axial fused PET/CT, corresponding PET and CT images of the brain (b-d, f-h) showed complete resolution of hypermetabolism involving mesial temporal lobes (arrowheads) and reduction in the 18F-FDG avidity in the basal ganglia and thalami. The coronal fused PET/CT and CT images of bilateral lungs (e,i) showed resolution of previously noted FDG avid ground glass haziness. The axial fused PET/CT, corresponding PET and CT images of the pelvic region (h-j), showed resolution of FDG avid soft tissue lesion with sclerosis in the ischial tuberosity (dashed arrows).

^{18}F-FDG avidity in the basal ganglia and thalami. The coronal fused PET/CT and CT images of bilateral lungs (e, i) showed resolution of previously noted FDG avid ground glass haziness. The axial fused PET/CT, corresponding PET and CT images of the pelvic region (h-j), showed resolution of FDG avid soft tissue lesion with sclerosis in the ischial tuberosity (dashed arrows).

Limbic encephalitis is the inflammation of the medial temporal lobe structures due to direct infectious or indirect autoimmune etiology.[1,2] Sarcoidosis is a rare cause of limbic encephalitis, has been reported by few authors.[3-5] The association has also been reported between limbic encephalitis and myasthenia gravis and sarcoidosis, the three different diseases with immune dysregulation as the common pathology.[6-10] This patient had a combination of myasthenia gravis, sarcoidosis, and limbic encephalitis, and ^{18}F-FDG PET/CT was helpful in the early identification of limbic encephalitis even before the onset of symptoms and the skeletal lesion. The knowledge about such rare combinations of autoimmune manifestations will be helpful for the better interpretation of the PET/CT and management of patients.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient (s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest

References

1. Tüzün E, Dalmau J. Limbic encephalitis and variants: Classification, diagnosis and treatment. Neurologist 2007;13:261-71.
2. Gultekin SH, Rosenfeld MR, Voltz R, Eichen J, Posner JB, Dalmau J. Paraneoplastic limbic encephalitis: Neurological symptoms, immunological findings and tumour association in 50 patients. Brain 2000;123:1481-94.
3. Douglas AC, Maloney AF. Sarcoidosis of the central nervous system. J Neurol Neurosurg Psychiatry 1973;36:1024-33.
4. Toudou-Daouda M, Assadeck H, Efared B. Sarcoidosis limbic encephalitis: A case report. Iran J Neurol 2017;16:156-8.
5. Richards P. Acute sarcoid meningo-encephalitis. Br Med J 1964;2:1576.
6. Monstad SE, Nøstbakken JK, Vedeler CA. CRMP5 antibodies found in a patient with limbic encephalitis and myasthenia gravis. J Neurol Neurosurg Psychiatry 2009;80:241-2.
7. Saper JR, Fry MB. Sarcoidosis presenting as a mediastinal mass in a patient with myasthenia gravis. Dis Nerv Syst 1977;38:57-9.
8. Andonopoulos AP, Papathanasopoulos PG, Karatza C, Angelopoulos J, Papapetropoulos T. Sarcoidosis in a patient with myasthenia gravis. Case report and review of the literature. Clin Rheumatol 1991;10:323-5.
9. De Bleecker J, van Aken L, van Landegem W, van den Abeele G, Cuvelier C. Myasthenia gravis and sarcoidosis: Report of 2 cases. Eur Neurol 1996;36:326-7.
10. Bonnet F, Dubuc J, Morlat P, Delbrel X, Doutre MS, de Witte S, *et al.* Sarcoidosis and comorbidity: Retrospective study of 32 cases. Rev Med Interne 2001;22:619-23.