Clinical Case Report

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Predictive value of brain ^{18}F-FDG PET/CT in macrophagic myofasciitis?

A case report

Axel Van Der Gucht, MD[a,*], Mukedaisi Abulizi, MD[a], Paul Blanc-Durand, MD[a], Mehdi Aoun-Sebaiti, MD[b,c], Berivan Emsen, MD[a], Romain K. Gherardi, MD, PhD[b,d,e], Antoine Verger, MD, PhD[f], François-Jérôme Authier, MD, PhD[b,d,e], Emmanuel Itti, MD, PhD[a,g]

Abstract

Rationale: Although several functional studies have demonstrated that positron emission tomography/computed tomography with ^{18}F-fluorodeoxyglucose (^{18}F-FDG PET/CT) appears to be efficient to identify a cerebral substrate in patients with known macrophagic myofasciitis (MMF), the predictive value of this imaging technique for MMF remains unclear.

Patient concerns: We presented data and images of a 46-year-old woman.

Diagnoses: The patient was referred to our center for suspected MMF due to diffuse arthromyalgias and cognitive disorder (involving an impairment of visual selective attention and a weakness in executive functions revealed by neuropsychological assessment) which occurred few years after last vaccine injections.

Interventions: After a first negative deltoid muscle biopsy, a brain ^{18}F-FDG PET/CT was performed and revealed the known spatial pattern of a cerebral glucose hypometabolism involving occipital cortex, medial temporal areas, and cerebellum.

Outcomes: Given the clinical suspicion of MMF and brain ^{18}F-FDG PET/CT findings, a 2nd deltoid muscle biopsy was performed and confirmed the diagnosis of MMF with typical histopathological features.

Lessons: This case highlights the predictive value of brain ^{18}F-FDG PET/CT as a noninvasive imaging tool for MMF diagnosis, even when muscle biopsy result comes back negative.

Abbreviations: ^{18}F-FDG = ^{18}F-fluorodeoxyglucose, MMF = macrophagic myofasciitis, PET/CT = positron emission tomography/computed tomography.

Keywords: aluminum hydroxide, brain imaging, ^{18}F-FDG, macrophagic myofasciitis, PET/CT

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[a] Department of Nuclear Medicine, H. Mondor Hospital, Assistance Publique-Hôpitaux de Paris/Paris-Est University, [b] INSERM U955-Team 10, [c] Department of Neurology, [d] Department of Pathology, H. Mondor Hospital, Assistance Publique-Hôpitaux de Paris/Paris-Est University, [e] Reference Center for Neuromuscular Disorders, H. Mondor Hospital, Assistance Publique-Hôpitaux de Paris, Créteil, [f] CHU Nancy, Nuclear Medecine and Nancyclotep Experimental Imaging Platform, Nancy, [g] INSERM U955-GRC Amyloid Research Institute, Créteil, France.

[*] Correspondence: Axel Van Der Gucht, Department of Nuclear Medicine, Hôpital Henri Mondor, Assistance Publique-Hôpitaux de Paris, Paris-Est University, 51 Ave. du Mal de Lattre de Tassigny, Créteil, France (e-mail: axel.vandergucht@gmail.com).

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1. Introduction

Macrophagic myofasciitis (MMF) is an unusual inflammatory myopathy characterized by specific muscle lesions. Lesions are assessing abnormal long-term persistence of aluminum hydroxide within macrophages at the site of previous vaccine injection containing aluminum hydroxide adjuvant particles. Evolution of this chronic disease is slow and symptoms (which typically include arthromyalgias, chronic fatigue, and a cognitive dysfunction) first may occur from months or years after the last vaccine injection.[1–5] Several positron emission tomography/computed tomography with ^{18}F-fluorodeoxyglucose (^{18}FFDG PET/CT) studies have investigated these cognitive disorders.[6,7] Although this imaging technique appears to be efficient to identify a cerebral substrate in patients with a known MMF, the predictive value of brain ^{18}F-FDG PET/CT to diagnose MMF remains unclear.

2. Case report

We presented the case of a 46-year-old woman which was referred to our center for suspected MMF due to chronic diffuse arthromyalgias, fatigue, and cognitive impairment which occurred at age 37, in the context of multiple aluminum hydroxide-based vaccines administration (hepatitis B vaccine at age 27, diphteria/tetanus/polio at age 32, 33, and 43). A first deltoid muscle biopsy was performed at age 45 and was normal

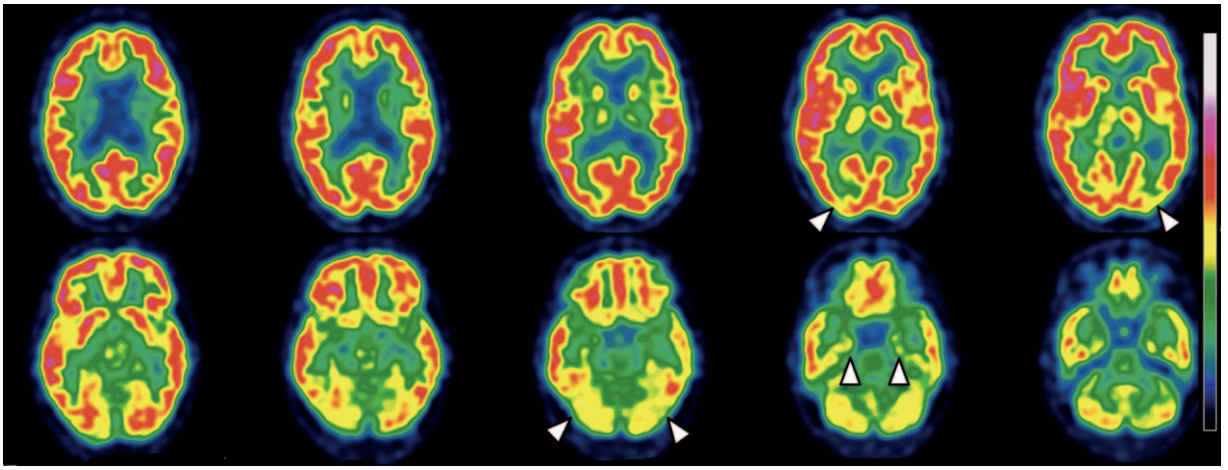

Figure 1. Brain ^{18}F-FDG PET is showed the known spatial pattern of a cerebral glucose hypometabolism involving occipital cortex, medial temporal areas, and cerebellum (white arrows). ^{8}F-FDG = ^{18}F-fluorodeoxyglucose, PET = positron emission tomography.

showing no inflammatory lesion. In spite of this result, the clinical probability of MMF was high, neurocognitive tests showing an impairment of visual selective attention and a weakness in executive functions. Therefore, a brain ^{18}F-FDG PET/CT was performed as part of the work-up on a Gemini GXL instrument PET/CT scanner (Philips, Da Best, The Netherlands) after intravenous injection of 265 MBq of ^{18}F-FDG. Informed consent was obtained. The patient was required to fast for at least 6 hours before undergoing the scan, had a normal blood sugar level, and an update neurosensory rest for 30 minutes. A low-dose helical CT was first performed for anatomical correlation and attenuation correction with the following parameters: X-ray tube tension

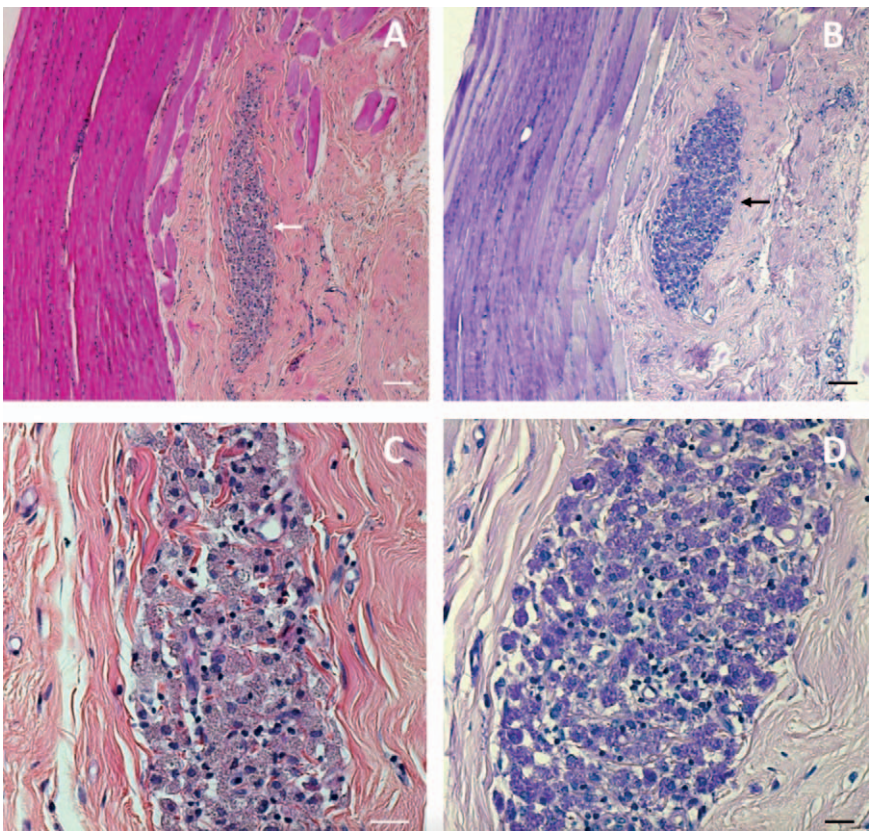

Figure 2. Histopathological findings: 2nd deltoid muscle biopsy. (A, B) MMF lesion: focal and well-circumscribed infiltration (arrow) cohesive large mononucleated cells in perimysium corresponding to macrophages intermingled with some lymphocytes, associated with fibrotic aspect of perilesional connective tissue. (C, D) Higher magnification showing the characteristic granular appearance of macrophages cytoplasm; no multinucleated giant cells were observed. PAS staining (B, D) showed strong positivity of MMF macrophages. Paraffin sections; hematoxylin-eosin (A, C) and PAS (B, D) staining; bars: 100 μm (A, B), 40 μm (C, D). MMF = macrophagic myofasciitis, PAS = Periodic Acid Schiff.

of 120 kV, current of 80 to 100 mAs, rotation time 0.5 seconds, pitch 0.938, and slice thickness 2 mm. Images were reconstructed using line of response-row action maximum likelihood algorithm (2 iterations, 28 subsets, and postfilter 5.1 mm), with and without CT attenuation correction (matrix size of 512×512, voxel size $4 \times 4 \times 4\,mm^3$). Visual analysis showed the known spatial pattern of a cerebral glucose hypometabolism involving occipital cortex, medial temporal areas, and cerebellum (Fig. 1).

Given the clinical suspicion of MMF and brain ^{18}F-FDG PET/CT findings, deltoid muscle biopsy was reiterated at age 46 and confirmed the diagnosis of MMF with typical histopathological features (Fig. 2).

3. Discussion

It is now well established that ^{18}F-FDG PET imaging, which measures glucose consumption in neuron bodies, is a highly useful imaging modality for the diagnosis of neurodegenerative disorders, in particular for specific types of dementia such as frontotemporal dementia, Alzheimer disease, dementia with Lewy bodies and more recently in MMF each of which has characteristics metabolic patterns.[8,9] In a large series of 100 patients, we described a peculiar spatial pattern of a cerebral glucose hypometabolism involving occipital lobes, temporal lobes, limbic system, cerebellum, and frontoparietal cortices and showed that MMF is a slowly progressive or nonprogressive disease, in accordance with the fact that neurologic symptoms – even if they fluctuate – do not worsen or improve over time.[10,11]

The teaching point of this report is that brain ^{18}F-FDG PET/CT should be performed in patients with suspected MMF and cognitive impairment. The risk of false-negative muscle biopsy is known, due to the heterogeneous and focal distribution of inflammatory sites.[12] Then, in case of suspected MMF associated with suggestive brain ^{18}F-FDG PET pattern, muscle biopsy at site of vaccine injections may have to be repeated after an initial negative result. This case highlights the predictive value of the brain ^{18}F-FDG PET/CT for MMF. Brain ^{18}F-FDG PET/CT could be considered as a noninvasive imaging tool to diagnose MMF even when muscle biopsy result comes back negative. Further studies are warranted to validate our findings.

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