The onset of the seizures are quite variable in CD as the duration of gluten exposure is important and a correlation can be found between the duration of the disease preceding the diagnosis (and treatment) and neurological findings as detected even years after the diagnosis. The two conditions may coexist for a prolong period of time before the actual clinical manifestations which may be till the late middle age as in our patient.

Our case demonstrate that neurological symptom such as epilepsy may be the first manifestation of CD and the diagnosis may be often challenging as the frequency of undiagnosed CD in neurological manifestations of unknown origin may be as high as 16%. A high index of suspicion for CD should be borne in mind in patients with epilepsy with nonspecific GI or constitutional symptoms. Although routine screening for CD in all patients with epilepsy is not cost-effective, it is reasonable to perform it in patients with neurological dysfunctions of unknown cause and in patients with intractable epilepsy. Early institution of a gluten-free diet in combination with antiepileptic treatment is beneficial as delay in diagnosis of CD in epilepsy patients or poor dietary compliance may adversely influence the overall outcome and lead to complications.

G. Das
P. Baglioni
Department of Diabetes and Endocrinology
Prince Charles Hospital
Mid Glamorgan
CF47 9DT, UK
email: drgdas@gmail.com

Follow-up of muscular sarcoidosis using fluorodeoxyglucose positron emission tomography

Sir,

Sarcoidosis is an immune-mediated condition affecting numerous organs, especially the lungs, lymph nodes, skin and eyes. Although asymptomatic muscle involvement is common in sarcoidosis (50–80% of histological cases), symptomatic myopathy is rare, being encountered in 0.5–2.3% of the patients. We recently observed a case of symptomatic myositis revealing a recurrence of sarcoidosis with favorable outcome after initiation of infliximab; our case is of particular interest, as it indicates that whole body fluorodeoxyglucose positron emission tomography (FDG-PET) is a useful test for both diagnosis and follow-up of myopathy in sarcoidosis.

A 43-year-old man was diagnosed as having both biopsy-proven pulmonary and muscle sarcoidosis in October 2005. In October 2008, the patient was...
asymptomatic; he still received combined therapy of prednisone (15 mg daily) and methotrexate (30 mg weekly), and steroid therapy regimen was reduced (12.5 mg per day). In January 2009, the patient was admitted for a 2-month history of myalgia and muscle weakness involving his lower limbs; he also complained of dry cough. On admission, general muscle strength was decreased; examination also revealed inspiratory fine crackles involving the lower lobes. Laboratory studies disclosed: erythrocyte sedimentation rate: 36 mm/h, C-reactive protein: 25 mg/l, creatine phosphokinase: 410 IU/l (n<130); renal and liver tests as well as blood protein electrophoresis were normal. Serum angiotensin-converting enzyme level was 176 IU/l (n<68). Computed tomography scan of the lungs showed bilateral hilar lymphadenopathy; pulmonary function tests revealed diffusing capacity for carbon monoxide to be 72% of predicted values without restrictive pattern (forced vital capacity: 103% of predicted values; vital capacity: 100% of predicted values). Whole-body PET demonstrated a marked uptake of FDG in the muscles of both calves and thighs (Figure 1), and enlarged hilar and mediastinal lymph nodes. As muscle involvement in sarcoidosis was refractory to steroids/immunosuppressive therapy, the patient was given anti-TNF-α: infliximab (5 mg/kg) at Weeks 0, 2, 6 and then 8 weekly. Infliximab therapy resulted in resolution of muscle clinical signs. At 6-month follow-up, the patient was symptom-free receiving prednisone (6 mg daily) and infliximab (5 mg/kg every 2 months); laboratory studies showed: creatine phosphokinase: 90 IU/l and decreased serum angiotensin-converting enzyme level (81 IU/l). Repeated FDG-PET further showed complete disappearance of: (i) FDG uptake in muscles of calves and thighs (Figure 2); and (ii) enlarged hilar and mediastinal lymph nodes.

Whole body FDG-PET is a promising method in sarcoidosis imaging, although it is still not incorporated in routine activity assessment of these patients. FDG-PET has, in fact, been reported to be helpful in identifying active sarcoid lesions, determined by intense F-18 FDG uptake in hilar and mediastinal lymph nodes and lung parenchyma.2–5 FDG-PET has also been found to accurately detect cardiac sarcoidosis, showing increased focal or diffuse pattern.2–5 Our case further underlines that FDG-PET is sensitive and non-invasive for the diagnosis of myopathy in patients with sarcoidosis; in this instance, FDG-PET indeed provided complete and detailed morpho-functional cartography of inflammatory

Figure 1. FDG-PET: marked uptake of F-18 FDG in the muscles of both calves and thighs.

Figure 2. Six-month follow-up FDG-PET: complete disappearance of FDG uptake in muscles of calves and thighs.
sarcoidosis-related active myositis. Furthermore, our case also suggests that FDG-PET is of value in monitoring the effectiveness of therapy in patients with both complex and multisystemic sarcoidosis, especially in the subgroup of patients with muscular sarcoidosis. In essence, we have interestingly observed a marked correlation between FDG-PET changes imaged and both clinical and biochemical sarcoidosis activity parameters; in turn, at 6-month follow-up, our patient was symptom-free and repeated FDG-PET showed complete disappearance of FDG uptake in muscles of calves and thighs. Nevertheless, no definite conclusion can be drawn from our latter findings, and our data warrant further investigations to assess the usefulness of FDG-PET in muscular sarcoidosis follow-up.

Our case is also original in that our patient with refractory muscular sarcoidosis was successfully given infliximab. Other investigators have previously suggested that anti-TNF-α may be an effective therapy for pulmonary and extrapulmonary sarcoidosis,6–10 although no definite conclusion can be drawn from these data. After immunosuppressive drugs failed to control muscular sarcoidosis in our patient, infliximab was used though anti-TNF-α agents have not yet been licensed for the therapy of sarcoidosis; however, because TNF-α has a pathological role in sarcoidosis, anti-TNF-α agents may be of therapeutic benefits in patients with sarcoidosis refractory to conventional drugs. Indeed, because TNF-α is recognized to be a factor in both initiation and maintenance of granulomas, anti-TNF-α antibody agents, which bind to and neutralize TNF-α, may result in inhibition of TNF-α after release from alveolar macrophages and other cells in patients exhibiting active sarcoidosis.6–8 Taken together, we therefore suggest that infliximab may be an effective therapy for sarcoidosis-related muscle involvement that is unresponsive to conventional therapy. Moreover, our patient did not develop adverse effects (e.g. infectious complication) related to infliximab.


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