

## F-18 FDG PET/CT in relapsing polychondritis

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**Abstract** We report a case of relapsing polychondritis for which fluorodeoxyglucose (FDG) positron-emission tomography/computed tomography (PET/CT) showed increased FDG accumulation in all rib cartilages, as well as in the larynx, trachea, and major bronchi. Contrast-enhanced CT during PET/CT showed smooth tracheal and bronchial wall thickening with calcification and airway narrowing. After steroid therapy, clinical symptoms and laboratory data were improved and cartilaginous FDG accumulation had completely disappeared. FDG PET/CT is considered to be a powerful radiological tool to assess the disease activity of relapsing polychondritis.

**Keywords** Relapsing polychondritis · F-18 FDG · PET/CT

### Introduction

Relapsing polychondritis is a rare multisystemic disease that is characterized by recurrent inflammation of the cartilaginous structures of the external ear, nose, peripheral joints, larynx, and tracheobronchial tree [1, 2]. Its cause is unknown, but it is thought to be autoimmune mediated. Airway involvement is present in up to 50% of patients

with relapsing polychondritis and is a major cause of morbidity and mortality [1, 2]. Diagnosis is made according to the empirically defined clinical criteria of McAdam et al. [1]. No specific histological finding is considered pathognomonic for relapsing polychondritis.

We report a case of fever and cough in which F-18 fluorodeoxyglucose (FDG) positron-emission tomography/computed tomography (PET/CT) depicted cartilage inflammation, which eventually led to a confirmed diagnosis of relapsing polychondritis.

### Case report

A 59-year-old woman presented with a 3-month history of persistent cough, sore throat, hoarseness, and low-grade fever. She also had pain at the sternocostal junctions and in the laryngeal region before admission. She had been previously diagnosed with nontuberculous (NTM) infection and was taking erythromycin. She also had a history of nasal polyp and chronic sinusitis, for which endoscopic sinus surgery had been performed. Physical examination revealed coarse crackles in the left back. The erythrocyte sedimentation rate (ESR) was 77 mm per h, and the C-reactive protein (CRP) level was 5.16 mg per deciliter (normal level <0.5). The white-cell count was 7.300 per mm<sup>3</sup>, with 78% neutrophils. All cultures, including NTM and serologic tests, were negative. The antinuclear factor level was normal.

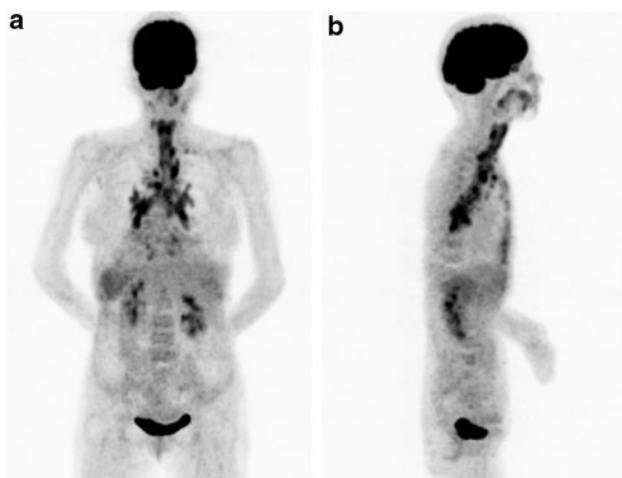
After admission, she was resistant to numerous antibiotics and antituberculous therapy and had a persistent fever of 38°C or higher.

Therefore, FDG PET/CT was performed to exclude malignancy and as a systemic search to explain fever of unknown origin. The patient fasted for 6 h before

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receiving an intravenous injection of 250 MBq F-18 FDG. Then, 90 min after the injection, attenuation correction CT (AC-CT) and emission scans were obtained from the upper thigh to the head using an integrated in-line PET/CT scanner (Biograph 16 LSO Hi-Rez; Siemens). Data acquisition began with AC-CT (with no contrast agent) using the standardized low-dose protocol of 120 kV, 50 mA, table speed of 18 mm, and pitch of 0.75 under shallow breathing; this was followed by PET with a 3-min emission acquisition time of 81 axial images at a 16.2-cm axial field of view per position. The images were reconstructed by the standard ordered-subset expectation maximization (OSEM) technique using 8 subsets and 3 iterations with a  $128 \times 128$  matrix for PET and  $512 \times 512$  matrix for CT. After PET data acquisition, contrast-enhanced CT from the neck to upper thighs was obtained 100 s after iopamidol 300 (Bayer Yakuhin, Osaka, Japan) injection at a rate of 2.0 ml/s. Contrast-enhanced CT was obtained using the standardized protocol of 120 kV, 200 mA, table speed of 18 mm, and pitch of 0.75 under breath holding. Maximum intensity projection (MIP) images of FDG PET showed increased FDG accumulation in all rib cartilages, as well as in the larynx, trachea, and major bronchi (Fig. 1a, b). The maximum standard uptake value ( $SUV_{max}$ ) of the respiratory tract at 90 min was 6.41. FDG also accumulated in enlarged cervical and mediastinal lymph nodes (Fig. 1a). Contrast-enhanced CT during PET/CT showed smooth tracheal and bronchial wall thickening with calcification and airway narrowing. This airway wall thickening was predominant in the anterior and lateral portions with sparing of the posterior membranous portion. In addition, CT demonstrated



**Fig. 1** Maximum intensity projection (MIP) images of FDG PET showed increased FDG accumulation in all rib cartilages, as well as in the larynx, trachea, and major bronchi (a, b). FDG was also accumulated in enlarged cervical and mediastinal lymph nodes (a)

multiple bilateral cervical, supraclavicular and mediastinal lymph node swelling with relatively homogeneous contrast enhancement (Fig. 2). On the basis of these FDG PET/CT findings, the diagnosis of relapsing polychondritis was suggested. She also met the criteria proposed by McAdam et al., because she also had neurosensory hearing loss and nonerosive polyarthritis.

The patient's condition improved with the administration of corticosteroids. Six months after starting therapy, the clinical symptoms had disappeared and laboratory data, such as CRP and ESR, had returned to normal levels. Cartilaginous FDG accumulation had completely disappeared on a second FDG PET/CT study (Fig. 3). In addition, contrast-enhanced CT showed improvement of the airway wall thickening and multiple lymph node swelling; however, the airway narrowing was still present at that time (Fig. 4).

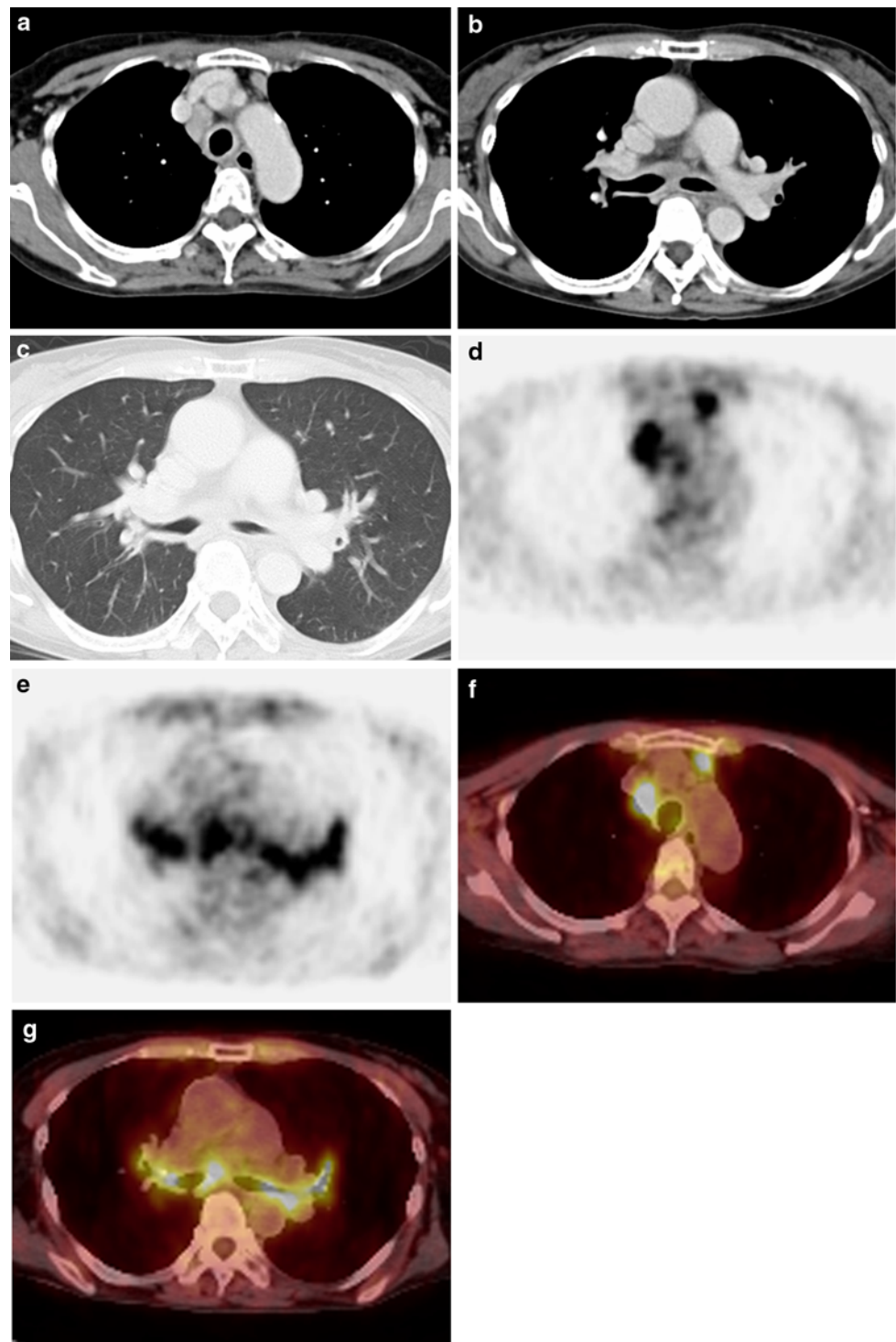
## Discussion

Relapsing polychondritis is considered very rare and the diagnosis may be challenging in the absence of typical auricular or nasal involvement [1, 2]. To date, laboratory data, symptoms and radiological findings have been used to assess relapsing polychondritis. In particular, assessment of respiratory tract involvement is important because it is a poor prognostic sign and a leading cause of death [1, 2]. Laboratory data, such as CRP and ESR, are useful to assess disease activity; however, it might reflect the inflammatory activity of other noncritical lesions instead.

Multidetector CT can visibly demonstrate the classic morphologic airway changes associated with relapsing polychondritis, including fixed airway narrowing and wall thickening with or without calcification [3–5]. The finding of smooth anterior and lateral airway wall thickening with sparing of the posterior membranous wall, as in our case, is virtually pathognomonic of relapsing polychondritis [3–5]. These changes are thought to occur secondary to cartilaginous destruction and fibrotic replacement, and thus reflect relatively late airway manifestations of relapsing polychondritis [4]. Identification of airway involvement in the initial stages of relapsing polychondritis is important because early aggressive treatment may potentially delay or prevent irreversible cartilaginous destruction. Lee et al. [5] reported that dynamic expiratory CT, which depicted functional airway abnormalities, including malacia and air trapping, may be the only airway abnormality identified during the early stages of relapsing polychondritis. This dynamic expiratory CT should be considered a routine component in the clinical suspicion of airway involvement.

On the other hand, wall thickening was improved after steroid therapy although airway stenosis was not improved

**Fig. 2** Contrast-enhanced CT during PET/CT showed smooth tracheal and bronchial wall thickening with calcification and airway narrowing. This airway wall thickening was predominant in the anterior and lateral sections with sparing of the posterior membranous section (a–c). Axial PET and fusion images showed moderate FDG accumulation in the trachea and bronchi, as well as in all rib cartilages (d–g). In addition, CT demonstrated multiple bilateral cervical, supraclavicular, and mediastinal lymph node swelling with relatively homogeneous contrast enhancement (a). These enlarged lymph nodes showed moderate FDG accumulation (d–g)



even after therapy in our case. It has been reported that stenosis may not necessarily improve even after inflammation is successfully suppressed [4, 5], making CT assessment after therapy difficult in some cases. In addition, reactive lymphadenopathy is considered to reflect the inflammatory activity in our case.

Radionuclide imaging has also been used to assess relapsing polychondritis [6–8]. Some case reports showed diffusely increased Tc-99m MDP uptake in the costochondral junction, and after prednisolone therapy, scintigraphic findings were improved [6, 7]; therefore, MDP scanning may be a valuable modality in the follow-up of

relapsing polychondritis. Okuyama et al. [8] reported the usefulness of gallium (Ga-67) scintigraphy in follow-up involving the subglottic region; however, with Tc-99m MDP bone scintigraphy it was difficult to depict respiratory tract involvement, and Ga-67 scintigraphy has poor spatial resolution and poor sensitivity.

Our case demonstrates the role of FDG PET/CT in the diagnosis and therapy response of relapsing polychondritis and shows the correlation between the findings on FDG PET/CT and disease activity, as assessed by clinical symptoms and laboratory data. Previously, only two case reports have been published concerning the usefulness of FDG PET in the diagnosis of relapsing polychondritis [9, 10]. Both case reports well depicted respiratory tract involvement, and the case reported by De Geeter and Vandecasteele [10] depicted rib cartilage FDG accumulation; however, neither report mentioned FDG accumulation in reactive lymphadenopathy, as was noted in our case. These findings suggest that FDG PET/CT is a useful

radiological tool to assess the early respiratory involvement of relapsing polychondritis and to assess disease activity with precise therapeutic responses. Moreover, by localizing the sites of active inflammation, FDG PET may guide the selection of a biopsy site.

In conclusion, FDG PET proved to be a useful radiological technique in the diagnosis and follow-up of relapsing polychondritis.

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**Fig. 3** MIP image of FDG PET after steroid therapy showed no cartilaginous FDG accumulation

**Fig. 4** Contrast-enhanced CT showed improvement of airway wall thickening and multiple lymph node swelling (a, b); however, airway narrowing was still present at the time

