Hong Kong J Radiol. 2023;26:202-5. Epub 16 Aug 2023 | https://doi.org/10.12809/hkjr2217627

CASE REPORT

Role of Fluorine-18 Fluorodeoxyglucose Positron Emission Tomography/Computed Tomography in Monitoring Relapsing Polychondritis: A Case Report

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INTRODUCTION

Relapsing polychondritis (RP) is a rare autoimmune inflammatory disease that affects cartilaginous tissue with consequent recurrent inflammation and deformation of the involved structures. Although the aetiology remains unknown, it is often associated with autoimmune disorders, with rheumatoid arthritis (RA) being the most common.[1,2] The sites that are first affected at disease onset include the auricular and nasal cartilages. Nonetheless other proteoglycan-rich structures including the eyes, heart valves and blood vessels can be involved. Due to its non-specific presentation and the lack of specific diagnostic methods, RP has a high risk of misdiagnosis, with a mean diagnostic delay of 2.9 years.[1]

There is no gold standard test or imaging to monitor RP. Although laboratory investigations such as erythrocyte sedimentation rate and C-reactive protein level can indicate active inflammation, they are neither sensitive nor specific.[2] Imaging modalities such as computed tomography (CT), although useful in the diagnosis of RP, are less sensitive for demonstrating the extent of active disease.[2] Histological confirmation is hindered by access difficulty and the associated complications of invasive procedures.[2] Recently, the use of fluorine-18 fluorodeoxyglucose positron emission tomography/CT (^{18}F-FDG PET/CT) has been investigated in the diagnosis, management and monitoring of RP. We present a 55-year-old man in whom ^{18}F-FDG PET/CT was used to assess RP.

CASE REPORT

A 55-year-old man with RA presented with a 3-year history of worsening shortness of breath and wheeze. The patient also experienced episodic nasal bridge pain and bilateral ear blockage. He had been treated with steroids (prednisolone) and immunosuppressants (sulphasalazine and azathioprine) for RA since 2018. In early 2021, he was admitted with increased shortness of breath, cough, and sputum. He had no chest pain or fever.

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Submitted: 11 Aug 2022; Accepted: 15 Nov 2022.

Contributors: Both authors designed the study, acquired the data, analysed the data, drafted the manuscript, and critically revised the manuscript for important intellectual content. Both authors had full access to the data, contributed to the study, approved the final version for publication, and take responsibility for its accuracy and integrity.

Conflicts of Interest: DWKC has disclosed no conflicts of interest. As an editor of the journal, EYPL was not involved in the peer review process.

Funding/Support: This study received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Data Availability: All data generated or analysed during the present study are available from the corresponding author on reasonable request.

Ethics Approval: This study was approved by the Institutional Review Board of The University of Hong Kong/Hospital Authority Hong Kong West Cluster, Hong Kong (Ref No.: HKWC-2022-026). The patient has provided written informed consent for all treatments, procedures, and publication.

Physical examination showed diffuse chest wheeze but was otherwise unremarkable. Chest X-ray showed no consolidation. Blood tests showed a normal white blood cell count (6.5×10^9/L) but elevated C-reactive protein level (14.4 mg/L).

CT of the thorax revealed smooth narrowing of the trachea and proximal bronchi and smooth tracheal wall thickening sparing the posterior wall. Changes were strongly suggestive of RP (Figure 1). A whole-body ^{18}F-FDG PET/CT confirmed the changes but with no corresponding increased uptake or hypermetabolic disease elsewhere (Figure 2a and 2b). A diagnosis of RP was made and the patient was treated with prednisolone, methotrexate, and mycophenolate mofetil.

The patient complained of unresolved bony pain with persistently elevated erythrocyte sedimentation rate (46 mm/h) and C-reactive protein level (71 mg/L). A follow-up ^{18}F-FDG PET/CT 12 months later to assess response to immunosuppressive therapy revealed new symmetrical uptake along the costochondral junctions (Figure 2c and 2d), suggestive of active RP. In view of the radiological findings and clinical progression, the dosage of methotrexate was increased; biologics would be considered if the disease remained refractory. The patient's symptoms subsequently improved with a decreasing trend of serum inflammatory markers.

DISCUSSION

The ^{18}F-FDG PET/CT was first reported in 2007 to provide metabolic information of an RP patient.[3] Since then, several case reports have shown ^{18}F-FDG PET/CT to be capable of determining organ involvement and evaluating disease activity and therapeutic response.[4-7]

The ^{18}F-FDG PET/CT findings in patients with RP comprise mainly airway wall thickening and calcification, airway stenosis and malacia, and air trapping.[1] The presence of symmetrically distributed high FDG-uptake lesions may also be diagnostic of RP.[2] Furthermore, ^{18}F-FDG PET/CT has a role in targeting biopsy sites, increasing remarkably the biopsy yield rate.[2] Nonetheless a retrospective study[8] found that in biopsy-proven auricular RP, or where there was tracheal involvement, the sensitivity and specificity of ^{18}F-FDG PET/CT were only 55.6% and 5.3%, respectively, raising questions about its usefulness in guiding biopsy.

A recent single-centre retrospective study compared ^{18}F-FDG PET/CT in patients before and after treatment and correlated the findings with clinical symptoms.[9] Follow-up ^{18}F-FDG PET/CT in most patients revealed a favourable treatment response, with significantly reduced visual scores and maximum standard uptake values in cartilaginous tissue. Another advantage of ^{18}F-FDG PET/CT lies in its ability to identify areas that are not clinically accessible but are of importance. In a patient with asymptomatic involvement of the aorta, ^{18}F-FDG PET/CT was able to identify increased FDG-uptake in the superior mesenteric artery and renal arteries.[9] Our case demonstrates the usefulness of ^{18}F-FDG PET/CT in assessing RP, providing objective evidence of

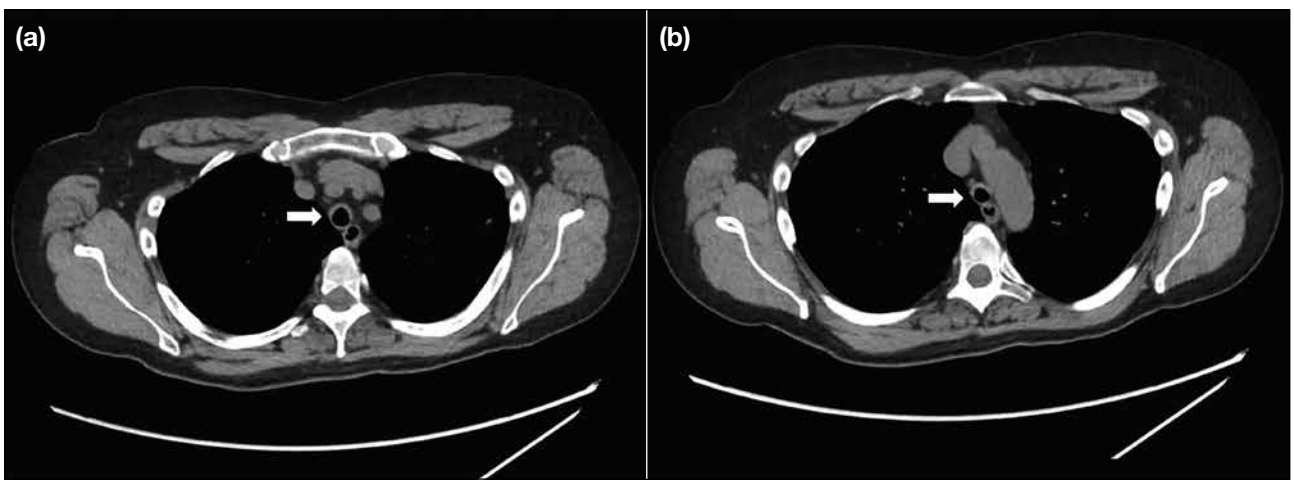

Figure 1. Axial non-contrast computed tomography images of the thorax showing (a) tracheal narrowing (arrow) and (b) smooth thickening of the tracheal wall sparing the posterior wall (arrow).

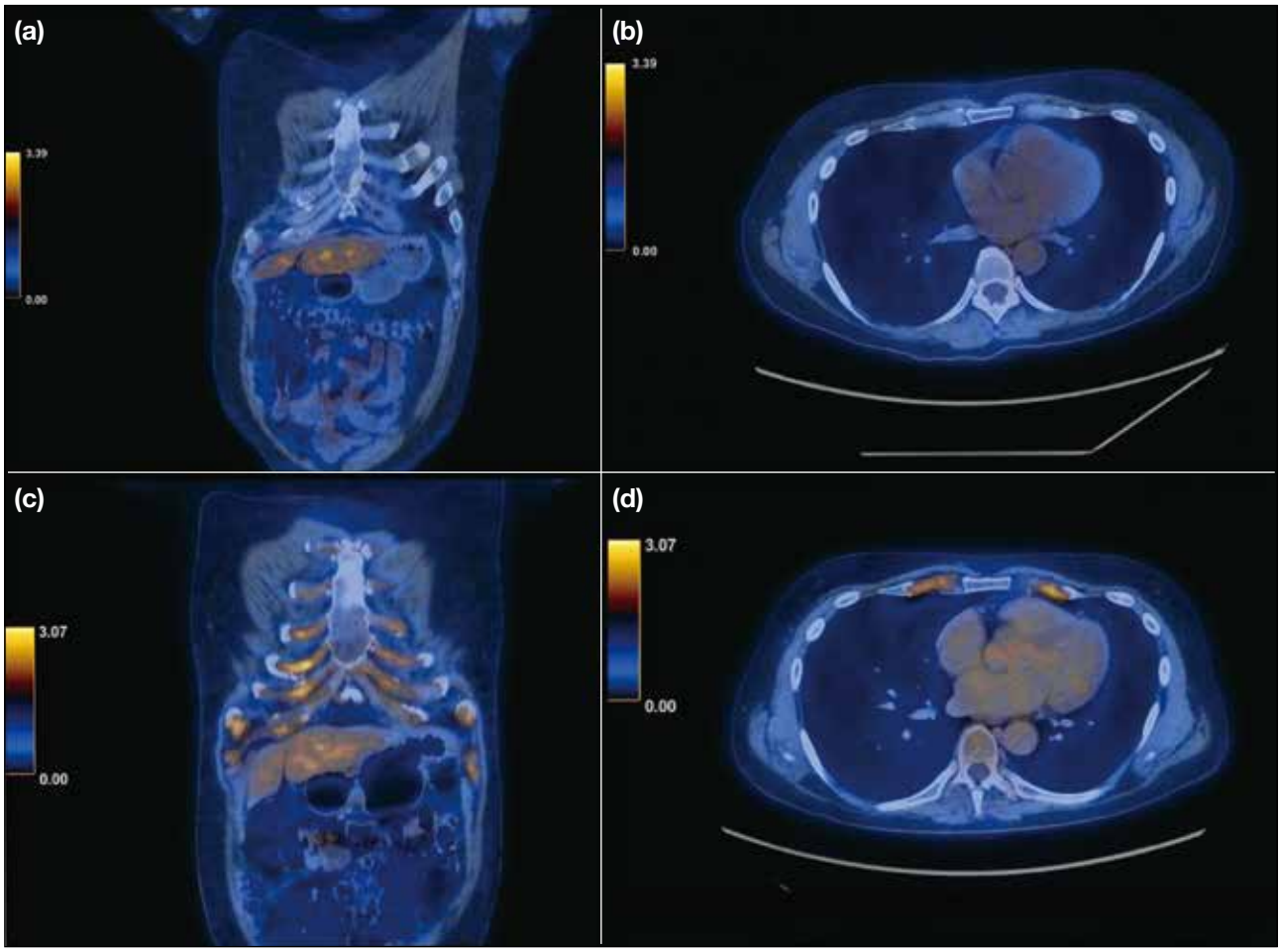

Figure 2. (a and b) Baseline fluorine-18 fluorodeoxyglucose positron emission tomography/computed tomography (^{18}F-FDG PET/CT). (a) Coronal fused ^{18}F-FDG PET/CT image shows no uptake along the costochondral junctions. (b) Axial fused ^{18}F-FDG PET/CT image shows no uptake along the costochondral junctions. (c and d) Follow-up ^{18}F-FDG PET/CT. (c) Coronal fused ^{18}F-FDG PET/CT image shows symmetrical uptake (maximum standard uptake value = 1.9-3.2) along the costochondral junctions. (d) Axial fused ^{18}F-FDG PET/CT image shows symmetrical uptake along the costochondral junctions.

active activity and prompting treatment escalation. This concurred with the clinical symptoms and serum inflammatory markers.

Apart from ^{18}F-FDG PET/CT, other imaging modalities have been investigated in the diagnosis and monitoring of RP. Although expiratory CT abnormalities are present in the majority of RP patients, only half demonstrate abnormalities on routine inspiratory CT scans.[10] Dynamic expiratory CT has been proposed as a routine diagnostic component if there is clinical suspicion of airway involvement.[10] In addition, studies have highlighted the use of bone scintigraphy using technetium-99m–methylene diphosphonate and gallium-67 citrate to evaluate disease activity and treatment response in RP patients.[11] In conclusion, ^{18}F-FDG PET/CT is useful in the follow-up of RP and in providing objective and measurable metrics to monitor disease activity.

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