



^{18}F -FDG PET/CT monitoring of non-tuberculous mycobacterial infection in a child with interleukin-12 receptor β -1 deficiency

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We report a 6-year-old girl with interleukin-12 receptor β -1 (IL-12R β 1) deficiency due to a homozygous deletion (g.1019_1020delAC) affecting codon His-339 in exon nine [1], and a mycobacterial infection by *M.genavense* with intestinal and mesenteric involvement, diagnosed by PCR in small bowel biopsy, with negative cultures and no antimicrobial susceptibility data.

She received empirical treatment with oral rifampicin, ethambutol, clarithromycin and levofloxacin for 4 months, which was later modified to oral rifampicin and clarithromycin plus intravenous ciprofloxacin and amikacin for 9 months, combined with IFN-gamma-1b (*Imukin*[®], Boehringer Ingelheim, Austria; initial dose of 50 mcg/m² three times/week subcutaneously, which was later increased to 80 mcg/m²), with partial improvement.

Persistence of active infection was confirmed by ^{18}F -fluorodeoxyglucose positron emission tomography/computed tomography (^{18}F -FDG-PET/CT) (Fig. 1). Treatment was changed to intravenous clarithromycin, amikacin, ciprofloxacin, linezolid and cefoxitin and IFN-gamma-1b dose was progressively increased (up to 250mcg/m² three times/

week), with significant clinical improvement after 3 months, confirmed by ^{18}F -FDG-PET/CT (Fig. 2).

During the following 24 months, treatment response was assessed using ^{18}F -FDG-PET/CT. PET/CT (Fig. 3), showing ongoing improvement that led to the progressive discontinuation of antimicrobial therapy after 36 months, with no further relapses reported in ^{18}F -FDG-PET/CT performed 7 months after finishing therapy (Fig. 4).

IL-12R β 1 deficiency predisposes to severe mycobacterial infections, with a mortality rate of up to 52% [2].

^{18}F -FDG PET/CT is a useful tool for monitoring therapeutic response in mycobacterial infections, especially in immunocompromised patients, disseminated disease, drug-resistant strains and unknown antimicrobial susceptibility. Serial ^{18}F -FDG PET/CT allows evaluation of disease activity and extent of the infection, facilitates adjustment of potentially toxic therapies and guides therapeutic decisions, especially when other clinical markers are suboptimal to assess disease activity [3–5].

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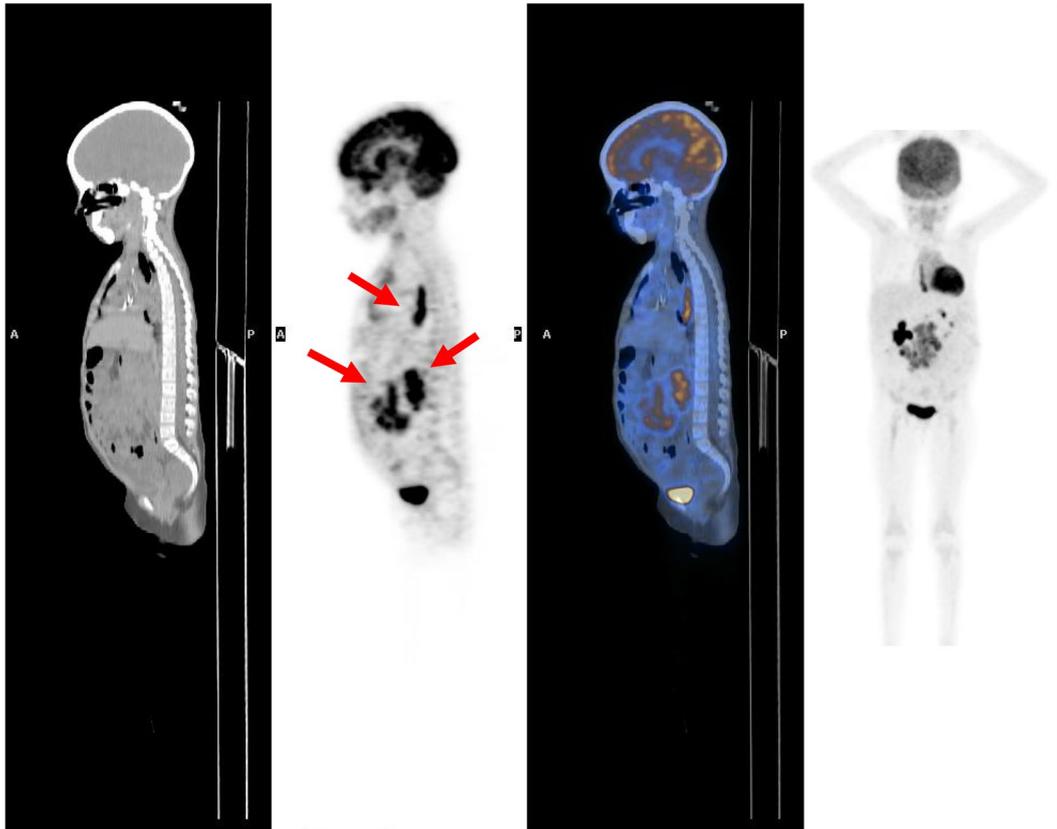


Fig. 1 First ^{18}F -FDG-PET/CT performed 9 months after starting treatment: Maximal intensity projection image (MIP) at the far right and sagittal views of the whole body from the ^{18}F -FDG PET (middle), corresponding low-dose non-contrast CT (left) and fused PET-

CT images (right) demonstrated active infection (arrows) with abnormal uptake in thoracic esophagus, abdominal mass in mesenteric and retroperitoneal pathological lymph nodes. Myocardial uptake is present as physiological uptake

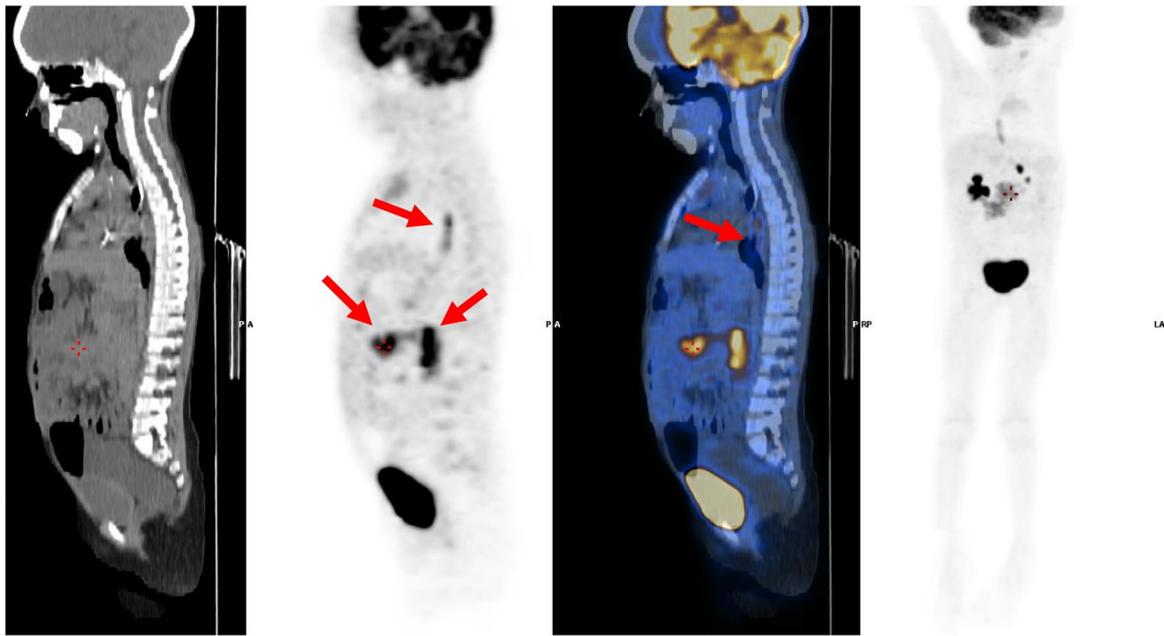


Fig. 2 ¹⁸F-FDG-PET/CT performed 3 months after the first one: Sagittal views of the whole body from the ¹⁸F-FDG PET (middle), corresponding low-dose non-contrast CT (left) and fused PET-CT

images (right), with important metabolic improvement after changes in antibiotic regimen. (arrows). Above all metabolic improvement can be seen in MIP image

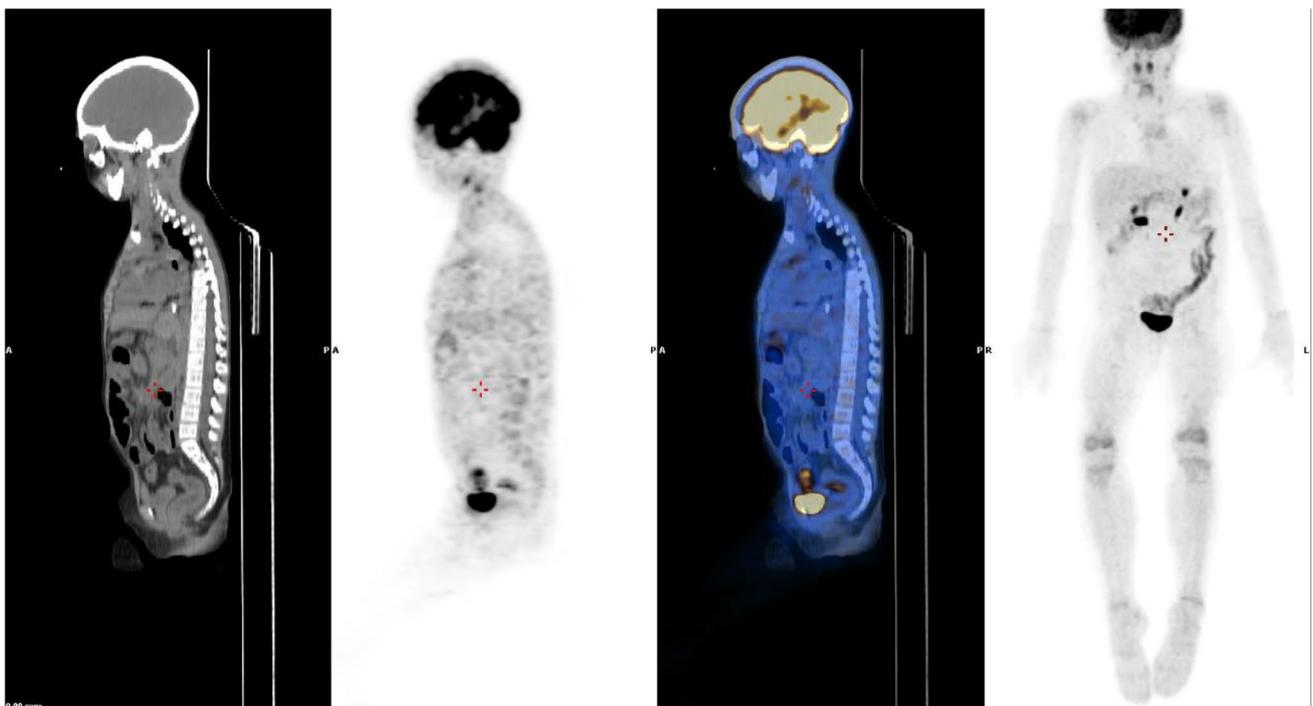


Fig. 3 ¹⁸F-FDG-PET/CT performed 24 months after the first one: sagittal views of the whole body from the ¹⁸F-FDG PET (middle), corresponding low-dose non-contrast CT (left) and fused PET-CT images (right), without signs of infection process

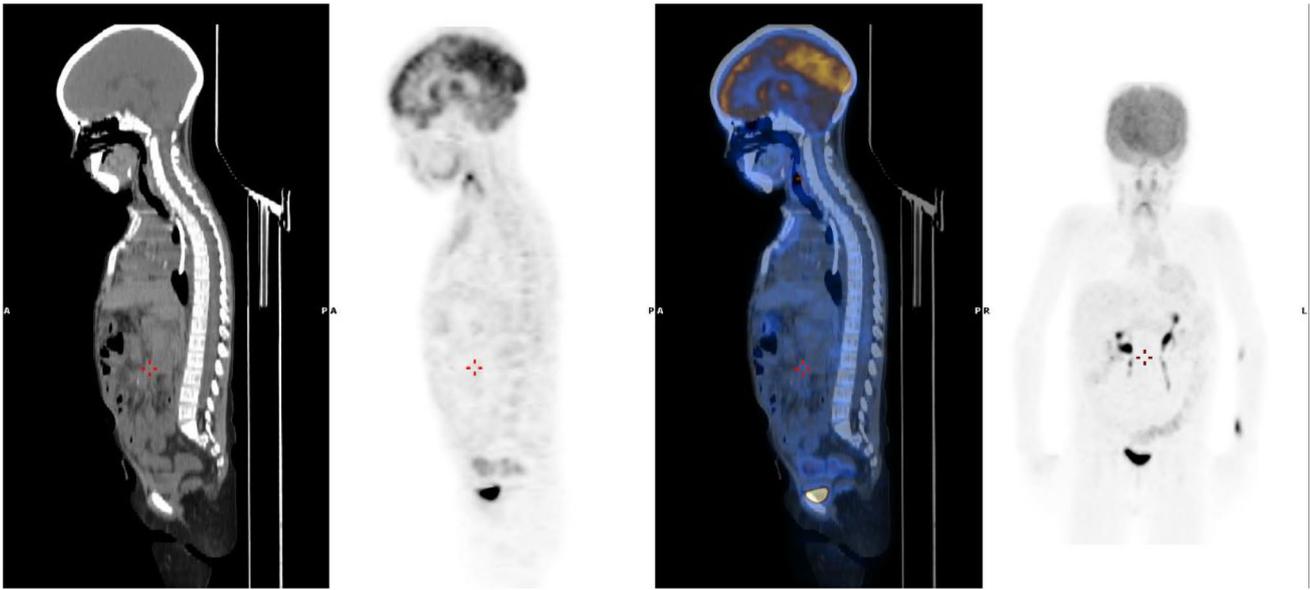


Fig. 4 18F-FDG-PET/CT performed 7 months after stopping the antibiotic therapy: sagittal views of the whole body from the 18F-FDG PET (middle), corresponding low-dose non-contrast CT (left) and fused PET-CT images (right), with no signs of infection process

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Compliance with ethical standards

Conflict of interest The authors indicate that they have no financial relationships relevant to this article or conflict of interest to disclose.

References

1. Gimenez-Sanchez F, Cobos-Carrascosa E, Sanchez-Forte M, Martinez-Lirola M, Lopez-Ruzafa E, Galera-Martinez R, et al. Different penetrance of disseminated infections caused by nontuberculous Mycobacteria in Mendelian susceptibility to mycobacterial disease associated with a novel mutation. *Pediatr Infect Dis J*. 2014;33:328–30.
2. de Beaucoudrey L, Samarina A, Bustamante J, et al. Revisiting human IL-12R β 1 deficiency: a survey of 141 patients from 30 countries. *Medicine (Baltimore)*. 2010;89:381–402.
3. Pelletier-Galarneau M, Martineau P, Zuckier LS, Pham X, Lambert R, Turpin S. 18F-FDG-PET/CT imaging of thoracic and extrathoracic tuberculosis in children. *Semin Nucl Med*. 2017;47:304–18.
4. Hofmeyr A, Lau WF, Slavin MA. Mycobacterium tuberculosis infection in patients with cancer, the role of 18-fluorodeoxyglucose positron emission tomography for diagnosis and monitoring treatment response. *Tuberculosis (Edinb)*. 2007;87:459–63.
5. Sato M, Hiyama T, Kaito K, Hayashi Y, Okumura T. Usefulness of F-18 FDG PET/CT in the assessment of disseminated *Mycobacterium avium complex* infection. *Ann Nucl Med*. 2009;23:757–62.