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Diffuse Bone Marrow Involvement of Langerhans Cell Histiocytosis Detected with F-18 FDG PET/CT

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What is known about this subject?

Importance of fluoro-2-deoxyglucose (FDG) positron emission tomography (PET) imaging in langerhans cell histiocytosis (LCH).

What this clinical images adds?

FDG PET could detect unknown involvement site of LCH.

Keywords: Langerhans cell histiocytosis, FDG, PET, bone marrow

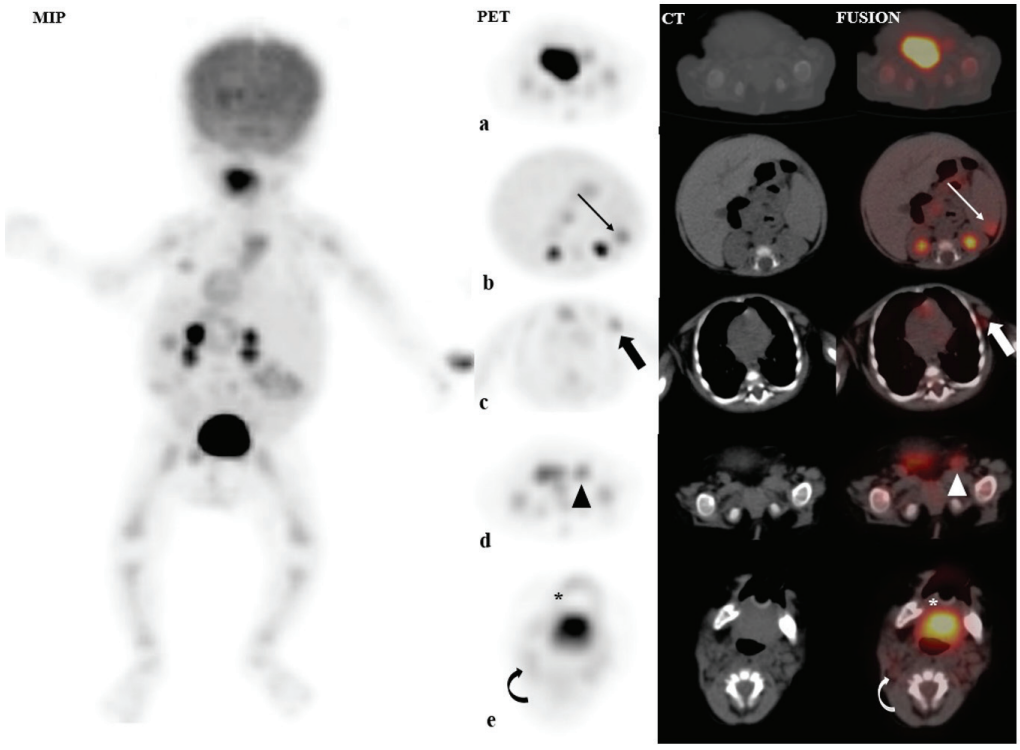

Figure 1. A four months-old girl infant presented with eczematous and squamates rash that began from the cranium and spread to the trunk in a day. Physical examination revealed disseminated erythematous, papules skin lesions. Blood test revealed only thrombocytopenia as pathologic (platelet:

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76 109 mg/dL; range: 247-580 109 mg/dL). Hemangiomatous suspected lesions were detected in the spleen, and liver with ultrasonography (USG). Therefore; F-18 fluoro-2-deoxyglucose positron emission tomography/computed tomography (F-18 FDG PET/CT) performed with Langerhans cell histiocytosis (LCH) prediagnosis. Maximum intensity projection image (left column) of PET demonstrates diffuse bone marrow FDG uptake, which is more prominent in the appendicular skeleton [maximum standardized uptake value (SUV_{max}): 1.4 g/mL]. Transaxial CT images (line a) do not reveal any abnormalities in the skeleton. Transaxial PET and fusion images show focal FDG uptake at the posterior aspect of the spleen (line b; thin arrow, SUV_{max}: 2.2) and mildly hypermetabolic enlarged lymph nodes in bilateral axillary (line c; thick arrow, SUV_{max}: 1.8), inguinal (line d; arrow head, SUV_{max}: 2.1), and cervical lymphatic stations (line e; curved arrow, SUV_{max}: 1.1). Because of crying during the uptake phase of radiopharmaceutical, intense FDG uptake was detected in the tongue base (line e; asterix, SUV_{max}: 6.5). Interestingly, no pathologic uptake was detected in skin lesions. USG confirmed pathologic axillary lymph nodes, but cervical lymph nodes were considered reactive. Skin punch biopsy revealed parakeratosis, and some horseshoe-shaped cells, some of which destroyed the dermoepidermal junction of the epidermis and formed nest up to the upper layers. Immunohistochemical staining was positive for S-100, CD1-a, langerin, CD48. Ki-67 was 5%. These findings were consistent with the diagnosis of LCH. LCH is a rare disease with an incidence of 4.6 cases per 1 million children under 15-years of age (1). Although bone involvement of LCH is common (2); bone marrow involvement of LCH is detected only in one-third of cases (3). Survival is poor in children with liver, spleen, or bone marrow involvement in LCH (4). Generally, conventional radiography was chosen as the first imaging modality. Magnetic resonance imaging (MRI), and diagnostic computed tomography are useful especially for identification of central nervous system and lung lesions (5). However, to evaluate the extent of the disease, and to monitor treatment response F-18 FDG PET/CT is a valuable imaging modality (6,7). Combined PET/MRI can improve sensitivity during primary staging (8). In this study, clinically unknown bone marrow involvement of LCH was detected with F-18 FDG PET/CT. Close observation of patients during the uptake phase of the radiopharmaceutical is critical to avoid false positive interpretation of PETs like tongue involvement in this study.

Ethics

Informed Consent: Informed consent for F-18 FDG PET scan was obtained from the case's parents.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: E.A., F.G., Concept: E.A., B.Y., Design: E.A., B.Y., Data Collection or Processing: E.A., F.G., B.Y., Analysis or Interpretation: E.A., B.Y., Literature Search: E.A., Writing: E.A., F.G.

Conflict of Interest: No conflict of interest was declared by the authors.

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REFERENCES

1. Guyot-Goubin A, Donadieu J, Barkaoui M, Bellec S, Thomas C, Clavel J. Descriptive epidemiology of childhood Langerhans cell histiocytosis in France, 2000-2004. Pediatr Blood Cancer 2008;51:71-75.
2. Reisi N, Raeissi P, Harati Khalilabad T, Moafi A. Unusual sites of bone involvement in Langerhans cell histiocytosis: a systematic review of the literature. Orphanet J Rare Dis 2021;16:1.
3. Minkov M, Pötschger U, Grois N, Gadner H, Dworzak MN. Bone marrow assessment in Langerhans cell histiocytosis. Pediatr Blood Cancer 2007;49:694-698.
4. Gadner H, Grois N, Pötschger U, et al. Improved outcome in multisystem Langerhans cell histiocytosis is associated with therapy intensification. Blood 2008;111:2556-2562.
5. Ferrell J, Sharp S, Kumar A, Jordan M, Picarsic J, Nelson A. Discrepancies between F-18-FDG PET/CT findings and conventional imaging in Langerhans cell histiocytosis. Pediatr Blood Cancer 2021;68:e28891.
6. Yadav D, Kumar R, Bal C. F-18 FDG PET/CT imaging in staging and response assessment in children with langerhans cell histiocytosis. J Nuc Med 2018;59:1604.
7. Albano D, Bosio G, Giubbini R, Bertagna F. Role of 18F-FDG PET/CT in patients affected by Langerhans cell histiocytosis. Jpn J Radiol 2017;35:574-583.
8. Mueller WP, Melzer HI, Schmid I, Coppenrath E, Bartenstein P, Pfluger T. The diagnostic value of 18F-FDG PET and MRI in paediatric histiocytosis. Eur J Nucl Med Mol Imaging 2013;40:356-363.