Interesting Image
Mol Imaging Radionucl Ther 2022;31:237-238 DOI:10.4274/mirt.galenos.2021.58672

Isolated Scapular Lesion of Langerhans Cell Histiocytosis Detected by ^{18}F-FDG PET/CT

^{18}F-FDG PET/BT ile Saptanan Langerhans Hücreli Histiyositozun İzole Skapula Lezyonu

Nuh Filizoğlu[1], Salih Özgüven[1], Hüseyin Kemal Türköz[2], Tunç Öneş[1], Halil Turgut Turoğlu[1], Tanju Yusuf Erdil[1]

[1]Marmara University, Pendik Training and Research Hospital, Clinic of Nuclear Medicine, İstanbul, Turkey
[2]Marmara University, Pendik Training and Research Hospital, Clinic of Pathology, İstanbul, Turkey

Abstract

Langerhans cell histiocytosis (LCH) is a rare disease that occurs mainly in pediatric patients and most adult LCH is considered a part of multisystem or multifocal disease. Only 7.3% of cases present as unifocal bone lesion. Herein, we present a case of an isolated scapular lesion of LCH in a 48-year-old man.

Keywords: Langerhans cell histiocytosis, ^{18}F-FDG PET/CT, bone

Öz

Langerhans hücreli histiyositoz (LHH), çoğunlukla pediatrik hastalarda ortaya çıkar ve yetişkin LHH genelikle multisistem veya multifokal hastalığın bir parçası olarak görülen nadir bir hastalıktır. Olguların sadece %7,3'ü tek odaklı kemik lezyonu olarak karşımıza çıkmaktadır.

Anahtar kelimeler: Langerhans hücreli histiyositoz, ^{18}F-FDG PET/BT, kemik

Address for Correspondence: Nuh Filizoğlu MD, Marmara University, Pendik Training and Research Hospital, Clinic of Nuclear Medicine, İstanbul, Turkey
Phone: + 90 216 396 86 48 **E-mail:** nuhfilizoglu@gmail.com ORCID ID: orcid.org/0000-0002-9139-9752
Received: 12.03.2021 **Accepted:** 22.07.2021

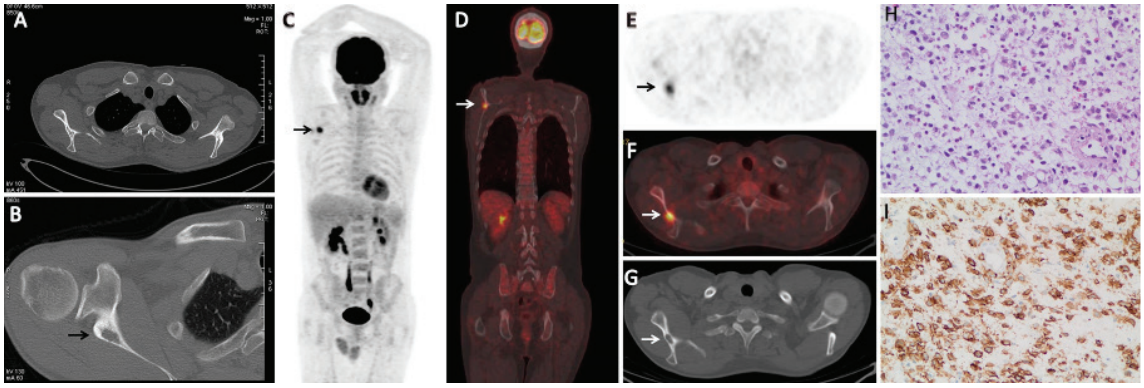

Figure 1. A 48-year-old man who was diagnosed with ankylosing spondylitis was admitted to a hospital for right shoulder pain. Computed tomography (CT) showed no lesions, which may cause the pain at shoulder area (A, transaxial CT image). Six months after his first CT, another CT was performed due to progression of his pain and revealed a lytic lesion in the right scapula (arrow in B, transaxial CT image). He was suspected to have scapular metastases or primary bone malignancies and was referred to ^{18}F-fluorodeoxyglucose (FDG) positron emission tomography (PET)/CT for further investigation. ^{18}F-FDG PET/CT showed 20x8 mm sized intense hypermetabolic [maximum standardized uptake value (SUV_{max}): 9.7] lytic lesion in the right scapula (arrows in C, coronal PET image; D, coronal fused PET/CT image; E, transaxial PET image; F, transaxial fused PET/CT image; G, transaxial CT image) and physiologic ^{18}F-FDG distribution on the rest of the body. Next a bone biopsy was performed. Histopathologic examination revealed neoplastic cells with vesicular nuclei, small nucleoli and nuclear grooves that were admixed with inflammatory cells including plasmacytes and eosinophils (H). Immunohistochemical examination of the tumor cells showed S100, tangerine and CD1a positivity (I). These findings were consistent with diagnosis of langerhans cell histiocytosis (LCH). LCH is a rare disease characterized by abnormal clonal proliferation and accumulation of pathological LC. Clinical presentation of LCH varies from isolated benign localization to multisystemic aggressive lesions (1). Although any organ can be affected, the most common site of involvement in LCH is skeletal. The accumulation of osteoclast-like multinucleated giant cells causes necrosis of the bone and consequently lead to formation of osteolytic lesions. The most common sites of involvement are the skull (27%) and involvement of other bones, such as the femur, humerus, spine, ribs and mandible, has also been reported. Isolated bony lesions of LCH are associated with a good prognosis and the lesion spontaneously regresses regardless of the type of treatment (2,3,4,5). Therefore, the most important aspect for treating LCH is categorizing the case correctly. ^{18}F-FDG PET/CT is superior at detecting new disease sites, recurrence, evaluating response to therapy, and distinguishing the metabolically active disease from inactive diseases to conventional imaging modalities (6). The utility of ^{18}F-FDG PET/CT for screening and follow-up in patients with LCH has been well established in the literature (7,8,9). LCH exhibits a relatively high SUV on ^{18}F-FDG PET/CT. Although the presence of scapular lesion as a part of the disseminated disease has been reported by prior publications, a case of LCH involving only the scapula has not been reported previously.

Ethics

Informed Consent: Obtained all appropriate patient consent forms.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: N.F., S.Ö., H.K.T., T.Ö., H.T.T., T.Y.E., Concept: N.F., S.Ö., Design: N.F., T.Y.E., Data Collection or Processing: N.F., S.Ö., Analysis or Interpretation: N.F., T.Y.E., Literature Search: N.F., S.Ö., Writing: N.F., S.Ö.

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

Financial Disclosure: The authors declared that this study received no financial support.

References

1. Monsereenusorn C, Rodriguez-Galindo C. Clinical characteristics and treatment of langerhans cell histiocytosis. Hematol Oncol Clin North Am 2015;29:853-873.
2. Agarwal KK, Seth R, Behra A, Jana M, Kumar R. 18F-fluorodeoxyglucose PET/CT in Langerhans cell histiocytosis: spectrum of manifestations. Jpn J Radiol 2016;34:267-276.
3. 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.
4. Chen L, Chen Z, Wang Y. Langerhans cell histiocytosis at L5 vertebra treated with en bloc vertebral resection: a case report. World J Surg Oncol 2018;16:96.
5. Sasaki H, Nagano S, Shimada H, Nakamura S, Setoguchi T, Komiya S. Clinical course of the bony lesion of single-system single-site Langerhans cell histiocytosis - is appropriate follow-up sufficient treatment? J Orthop Sci 2018;23:168-173.
6. 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.
7. Nguyen BD, Roarke MC, Chivers SF. Multifocal Langerhans cell histiocytosis with infiltrative pelvic lesions: PET/CT imaging. Clin Nucl Med 2010;35:824-826.
8. Obert J, Vercellino L, Van Der Gucht A, de Margerie-Mellon C, Bugnet E, Chevret S, Lorillon G, Tazi A. 18F-fluorodeoxyglucose positron emission tomography-computed tomography in the management of adult multisystem Langerhans cell histiocytosis. Eur J Nucl Med Mol Imaging 2017;44:598-610.
9. Lee HJ, Ahn BC, Lee SW, Lee J. The usefulness of F-18 fluorodeoxyglucose positron emission tomography/computed tomography in patients with Langerhans cell histiocytosis. Ann Nucl Med 2012;26:730-737.