OPEN ACCESS

Diffuse ^{18}F -fluorodeoxyglucose accumulation in the bone marrow of a patient with haemophagocytic lymphohistiocytosis due to Hodgkin lymphoma

Shu Harada,¹ Tsutomu Shinohara,² Keishi Naruse,³ Hisanori Machida¹

¹Division of Pulmonary Medicine, National Hospital Organization Kochi Hospital, Kochi, Japan

²Department of Clinical Investigation, National Hospital Organization Kochi Hospital, Kochi, Japan

³Division of Pathology, National Hospital Organization Kochi Hospital, Kochi, Japan

Correspondence to
Dr Tsutomu Shinohara,
shinoharat@kochi2.hosp.go.jp

Accepted 15 October 2016

DESCRIPTION

A 53-year-old man was referred to our hospital due to malaise with intermittent high fever. Thoracoabdominal CT scan revealed abdominal lymphadenopathy with splenomegaly. Hepatic disorder (aspartate transaminase 47 IU/L; alanine transaminase 53 IU/L), peripheral blood cytopenia (red blood cell count $370 \times 10^4/\mu\text{L}$; haemoglobin 11.0 g/dL; haematocrit 32.9%; platelet count $7.9 \times 10^4/\mu\text{L}$), hypertriglyceridaemia (184 mg/dL), hyperferritinaemia (4111 ng/mL) and an increased serum soluble interleukin-2 receptor level (4450 U/mL) were observed. Bone marrow (BM) aspiration showed erythrocyte phagocytosis by macrophages (figure 1A). However, no atypical cells were detected. Additional BM trephine biopsy was not performed. Pathological examination of a laparoscopic lymph node biopsy revealed Reed-Sternberg cells (figure 1B), and atypical cells were positive for CD30 but not for CD3, CD20 or CD79a indicating nodular sclerosis Hodgkin lymphoma (HL). A diagnosis of lymphoma-associated haemophagocytic lymphohistiocytosis (LA-HLH) was established. ^{18}F -fluorodeoxyglucose positron emission tomography (FDG-PET) performed for the staging showed splenic and multiple abdominal lymph node lesions and diffuse accumulation within bones such as the vertebrae and pelvis (figure 2A). After four courses of ABVD (doxorubicin, bleomycin, vinblastine and dacarbazine) chemotherapy, FDG-PET showed the disappearance of all abnormal accumulations (figure 2B). Involved-field

Figure 2 ^{18}F -fluorodeoxyglucose positron emission tomography before (A) and after (B) the four courses of ABVD (doxorubicin, bleomycin, vinblastine and dacarbazine) chemotherapy. (A) Splenic (arrow) and multiple abdominal lymph node (arrowheads) lesions and diffuse accumulation within bones such as vertebrae and pelvis. (B) Disappearance of all abnormal accumulations.

radiation therapy for abdominal lesions was sequentially added.

Patients with HLH with FDG-PET images indicating multiple lymphadenopathy and/or patchy multiple bone lesions may appear to be LA-HLH with BM involvement (BMI).¹ However, in comparison with BM examinations, FDG-PET can frequently show false-positive for the evaluation of BMI in patients with LA-HLH because HLH can induce diffuse hypermetabolism in BM reflecting a systemic cytokine storm.² Previously, several cases

Figure 1 Pathological findings. (A) Bone marrow aspirate smear showing macrophages laden with erythrocytes (arrow). (B) Abdominal lymph node biopsy showing scattered large atypical multinucleated cells (Reed-Sternberg cells; arrows) in prominent small lymphocytes.

CrossMark

To cite: Harada S, Shinohara T, Naruse K, et al. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2016-217555

Images in...

of HLH due to non-HL with diffuse FDG accumulation in BM were reported.²⁻³ Since we performed BM aspiration only without BM trephine biopsy, skeletal and/or BM HL involvement cannot be fully ruled out. However, there is a possibility that HLH due to HL also can induce diffuse hypermetabolism in BM.

Learning points

- ▶ ¹⁸F-fluorodeoxyglucose positron emission tomography (FDG-PET) has become a standard tool for the initial staging and reassessment of Hodgkin lymphoma, which is generally characterised by contiguous lymph node involvement.
- ▶ Haemophagocytic lymphohistiocytosis, which is the uncontrolled activation of lymphocytes and macrophages caused by various diseases including malignant lymphoma, can induce diffuse hypermetabolism in the bone marrow reflecting a systemic cytokine storm.
- ▶ Recognition of false-positive findings on FDG-PET is necessary for the evaluation of bone marrow involvement in order to accurately determine the stage of Hodgkin lymphoma, which requires bone marrow examination.

Contributors SH drafted the initial manuscript. TS edited and submitted the manuscript. SH, TS and HM were involved in diagnosing and treating the patient. KN performed pathological studies.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

Open Access This is an Open Access article distributed in accordance with the terms of the Creative Commons Attribution (CC BY 4.0) license, which permits others to distribute, remix, adapt and build upon this work, for commercial use, provided the original work is properly cited. See: <http://creativecommons.org/licenses/by/4.0/>

REFERENCES

- 1 Wang J, Wang D, Zhang Q, *et al.* The significance of pre-therapeutic F-18-FDG PET-CT in lymphoma-associated hemophagocytic lymphohistiocytosis when pathological evidence is unavailable. *J Cancer Res Clin Oncol* 2016;142:859–71.
- 2 Yang YQ, Ding CY, Xu J, *et al.* Exploring the role of bone marrow increased FDG uptake on PET/CT in patients with lymphoma-associated hemophagocytic lymphohistiocytosis: a reflection of bone marrow involvement or cytokine storm? *Leuk Lymphoma* 2015;19:1–8.
- 3 Yiu CR, Kao YH, Phipps C, *et al.* Positron emission tomography findings in patients with lymphoma-associated haemophagocytic syndrome. *Singapore Med J* 2011;52:e156.

Copyright 2016 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.
BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- ▶ Submit as many cases as you like
- ▶ Enjoy fast sympathetic peer review and rapid publication of accepted articles
- ▶ Access all the published articles
- ▶ Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow