

Kikuchi-Fujimoto Disease with 18F-fludeoxyglucose Uptake in Cervical Lymph Nodes on Dual-time-point Imaging Positron Emission Tomography/computed Tomography Mimicking Malignant Disease

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Abstract

Kikuchi-Fujimoto disease (KFD) is a benign but self-limiting disorder. However, KFD is often misdiagnosed as a malignant disease. Although 18F-fludeoxyglucose (FDG) uptake on dual-time-point imaging (DTPI) positron emission tomography (PET)/computed tomography (CT) is useful in distinguishing malignant from benign disease, the latter sometimes mimics malignancy on DTPI PET/CT, resulting in a misdiagnosis. Here, we describe the case of a 30-year-old woman who complained of cervical lymphadenopathy. PET showed increased FDG uptake in multiple lymph nodes, with a maximum standardized uptake value (SUVmax) of 19.0 in the early phase to 21.8 in the late phase. A biopsy was performed, and pathological examination revealed KFD. KFD with FDG uptake in lymph nodes on DTPI PET/CT is rare and difficult to be distinguished from a malignant disease.

Keywords: Cervical lymphadenopathy, dual-time-point imaging, fludeoxyglucose-positron emission tomography/computed tomography, Kikuchi-Fujimoto disease

Introduction

Kikuchi-Fujimoto disease (KFD), known as histiocytic necrotizing lymphadenitis, is a benign but self-limiting disorder. KFD often shows with symptoms similar to those of malignant disease and is hence difficult to distinguish from malignancy.^[1] Thus, imaging studies are often used to confirm the diagnosis. Recently, several studies have shown that dual-time-point imaging (DTPI) 18F-fludeoxyglucose (FDG) positron emission tomography (PET)/computed tomography (CT) may be useful in differentiating malignancy from benign

disease in lymph nodes.^[2-7] However, this differentiation is not always possible. Here, we describe a case of KFD in which FDG uptake in cervical lymph nodes on DTPI PET/CT mimicked malignant lesions.

Case Report

Patient history and background

A 30-year-old woman was referred to an oral and maxillofacial surgeon because of right submandibular lymphadenopathy and fatigue. She reported a fever with body temperature of up to 38.0°C daily for nearly 2 weeks, without upper respiratory symptoms. The fever

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had developed after the extraction of the right wisdom tooth, and she denied any foreign travel prior to the symptoms.

Physical examination

Slight facial swelling and anterior cervical lymphadenopathy with tenderness was observed, and wound healing was noted in the extraction socket.

Laboratory tests

The patient had a slightly elevated white blood cell count (8,800 cells/mm²; neutrophils, 88%; lymphocytes, 3%; atypical lymphocytes, 1%) and C-reactive protein level (1.32 mg/dL), with normal lactate dehydrogenase and alkaline phosphatase levels. Serum levels of immunoglobulin (Ig) G, IgA, IgM, IgE, complement components (C) 3, C4, carcinoembryonic antigen, and alpha-fetoprotein were within normal ranges. The patient tested negative on the tuberculin-purified protein derivative test and on tests for cytomegalovirus, Epstein-Barr virus, Herpes simplex virus, and toxoplasma. Tests for extractable nuclear antigen and antinuclear antibody were negative as well.

Mild bacterial infection of the extraction socket was suspected and antibiotic therapy was initiated. However, the patient showed no improvement after 15 days. Accordingly, contrast-enhanced neck CT was performed, that showed cervical lymph node enlargement, with maximum dimensions of 19.0 mm × 19.0 mm [Figure 1].

Under the suspicion of malignancy, whole-body FDG PET/CT was performed. The early and late phase images were obtained 60 min and 120 min after the injection of FDG, respectively. The PET scan showed multiple lymph nodes with increased FDG uptake in the right cervical area, excluding the extraction socket. The maximum standardized uptake value (SUV_{max}) of the lesion was confirmed with dual-phase scanning to be 19.0 in the early phase to 21.8 in the late phase [Figure 2]. These findings suggested malignant disease such as lymphoma.

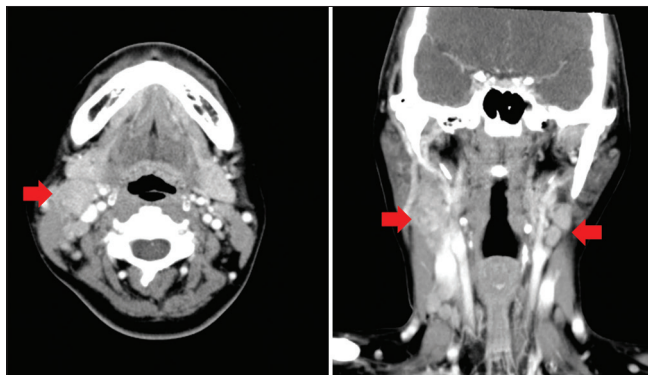


Figure 1: Enhanced neck computed tomography image showing cervical lymph node enlargement (arrows). The maximum dimensions were 19.0 mm × 19.0 mm

A biopsy was performed, but histological examination revealed KFD, with no evidence of lymphoma.

Because KFD is a benign condition, the patient was placed under observation and no therapy was administered. Cervical lymphadenopathy disappeared after 3 months.

No recurrence has been noted for 2.5 years since diagnosis.

Discussion

KFD, also known as histiocytic necrotizing lymphadenitis, was first described in Japan in 1972. It is a benign but self-limiting disorder.^[1] The most common manifestation of KFD is cervical lymphadenopathy with fever and night sweats, and laboratory data are not specific.^[8] The etiology of KFD remains unknown, but KFD is thought to be triggered by an autoimmune or viral process. Usually, KFD is limited to the cervical area, but it may, in addition, involve lymph nodes in the whole body. Surgical excision is necessary to make a definitive diagnosis.^[1]

In the present case, physical examination findings were nonspecific. In addition, although no abnormal findings were noted in the extraction socket, routine laboratory data indicated mild inflammation and no viral infection. Further, there was no evidence of scleral icterus or hepatosplenomegaly. Accordingly, malignant disease was suspected, and FDG PET/CT was consequently performed.

FDG PET/CT is a widely used imaging modality in oncology. In particular, DTPI FDG PET/CT has been shown in multiple studies to be useful in differentiating malignancy from benign conditions.^[2-8] However, differentiation with DTPI FDG PET/CT is not always possible, resulting in a misdiagnosis due to a benign condition mimicking a malignant disease. Generally, DTPI is performed at 1 h and 2 h after FDG injection,

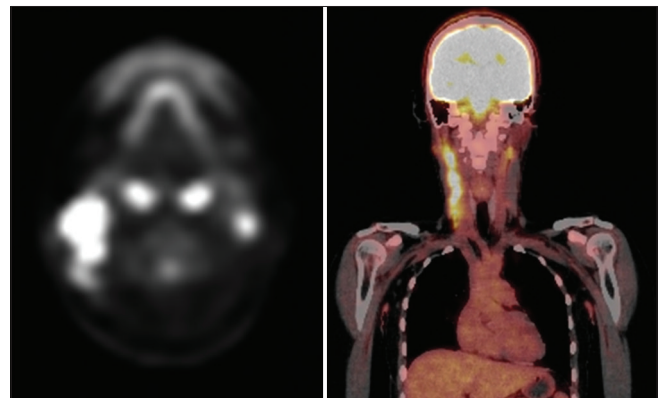


Figure 2: Delayed phase 18F-fludeoxyglucose positron emission tomography/computed tomography imaging. Multiple swollen lymph nodes were detected in the right cervical region. The maximum standardized uptake value was 21.8 in the late phase

respectively. The criterion for the diagnosis of malignancy on delayed imaging is an SUV_{max} of more than 2.5, an increased SUV Δ SUV on DTPI compared to the initial images, or both.^[8] In addition, some studies have used a retention index (RI) of >0% or 10% as the preferred indicator of malignancy, with RI calculated as follows: RI (%) = 100% × [SUV (delayed) - SUV (initial)]/SUV (initial).^[9-11] However, active granulomatous lesions and inflammation or acute infection sometimes show high FDG (delayed) uptake on DTPI, similar to the case for malignant lesions.^[10] One previously reported case of KFD was associated with high FDG (delayed) activity (SUV_{max} of 13.5 to 21.1 in the cervical region).^[12] Another study reported that surgical biopsy is necessary for the differentiation of malignant disease from KFD, regardless of DTPI findings.^[13] In agreement with this case, DTPI was not useful in distinguishing KFD from malignant disease in our case as well; lymphoma was suspected to be based on the DTPI findings. We found that FDG PET/CT showed an SUV_{max} of 19.0 to 21.8 and increased SUV (delayed) activity [Δ SUV = 2.8 (>2.5) and RI = 14.7% (>10%)]. Thus, lymphoma was suspected, and a biopsy was performed. Based on our experience with this case, further investigations are required to diagnose KFD in cases of lymph node uptake on DTPI.

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Conflicts of interest

There are no conflicts of interest.

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