Clinical Case Report

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Castleman disease in the hilum liver mimicking the lymph node metastasis of hepatocellular carcinoma on ^{18}F-FDG PET/CT

A case report

Yan Hu, MD[a,b], Yalan Liu, MD[a,c], Hongcheng Shi, MD, PhD[a,b,*]

Abstract

Rationale: Castleman disease is a rare lymphoproliferative disorder which etiology is unknown. It is divided into 2 subtypes: the plasma cell variant and the hyaline vascular variant. The most common site is the hilum of the lungs and mediastinum. Castleman disease is often ignored by clinicians because of not frequently encountering in clinical and the lower uptake of ^{18}F-FDG. Here, we report a case of hepatocellular carcinoma accompanied by Castleman disease in the hilum of the liver, which was misdiagnosed as the lymph node metastasis in ^{18}F-FDG PET/CT imaging.

Patient concerns: A 63-year-old male with chief complaint of acratia and poor appetite for 2 weeks.

Diagnoses: Hepatocellular carcinoma accompanied by Castleman disease in the hilum of liver.

Interventions: Right hemihepatectomy and hilar lymph node dissection was performed and preventative transcatheter arterial chemoembolization therapy was given after the surgery.

Outcomes: The case had multiple systemic metastasis due to tumor progression.

Lessons: Castleman disease is a benign and rare lympholiferative disease, and easy to be ignored in clinical. The case of hepatic carcinoma with Castleman disease in the hilum of liver has never been reported before. Here, we report it and hope it provides a differential diagnosis for clinicians in the following similar cases.

Abbreviations: ^{18}F-FDG = fluorine-18-fluoro-2-deoxyglucose, AFP = alpha-fetoprotein, CA-199 = carbohydrate antigen 19-9, CEA = carcinoembryonic antigen, PET/CT = positron emission tomography/computed tomography, SUV = standardized uptake value.

Keywords: Castleman disease, hepatocellular carcinoma, PET/CT

1. Introduction

Castleman disease is a low-grade atypical lymphoproliferative disorder mostly located in the mediastinum and hilum of the lungs.[1–4] Hyaline vascular and plasma cell variant are 2 histological subtypes, the type of former is usually asymptomatic and presented as a mass and the later type often characterized by systematic symptoms like fever, anemia, weight loss, night sweat and generalized lymphadenopathy.[5–7] Castleman disease often shows mild to moderate ^{18}F-FDG uptake, which is well correlated with disease multicentricity and clinical manifestation.[8,9] Castleman disease can be occurred in the region of lymph nodes in the whole body, but it is rare located in the liver or the hilum of liver. Here, we report a case of hepatocellular carcinoma in the liver accompanied by the Castleman disease in the hilus hepatis, which was misdiagnosed as the lymph node metastasis in ^{18}F-FDG PET/CT imaging.

2. Case report

A 63-year-old male with chief complaint of acratia and poor appetite for 2 weeks was suggested liver cancer with heaptic hilum and retroperitoneum lymphadenectasis by abdominal magnetic resonance imaging in other hospital. He was referred to our hospital for further diagnosis and treatment. Hepatitis B markers of HBsAg, HBsAb, HBeAg, HBeAb, and HBcAb were negative. Tumor markers of AFP, CEA, and CA-199 were 68.0, 1.4, and 4.3 ng/mL. Routine physical examination was normal. ^{18}F-FDG PET/CT scanning demonstrated a low-density lesion in the right lobe of liver, the maximum cross section was approximately 50.8 × 39.8 mm and the margin was ill-defined. The abnormal increase of glucose metabolism was heterogeneous, and the SUVmax was about 17.8 (Fig. 1B–D). Heaptic hilum region appeared multiple swollen lymph nodes, the maximum cross section of the largest lymph node in the hepatic hilum was about 13.2 × 9.8 mm and the SUVmax was 1.8

Editor: N/A.

Informed Consent: The patient gave the informed consent.

The authors report no conflicts of interest.

[a] Department of Nuclear Medicine, [b] Institute of Nuclear Medicine, [c] Department of Pathology, Zhongshan Hospital, Fudan University, Shanghai, P.R. China.

[*] Correspondence: Hongcheng Shi, Department of Nuclear Medicine, Zhongshan Hospital, Fudan University, 180, Fenglin Road, Shanghai, P.R. China (e-mail: shihongcheng163@163.com).

Medicine (2018) 97:12(e9939)

Received: 15 December 2017 / Accepted: 30 January 2018

http://dx.doi.org/10.1097/MD.0000000000009939

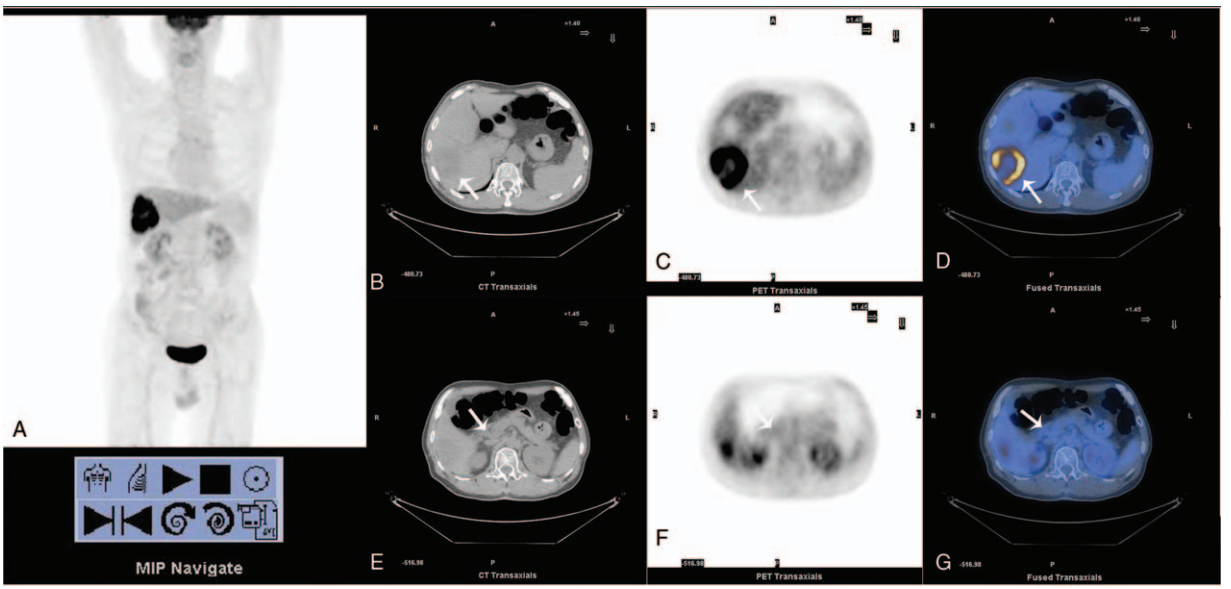

Figure 1. ^{18}F-FDG PET/CT scanning demonstrated a low density lesion in the right lobe of liver, the maximum cross section was approximately 50.8 × 39.8 mm and the SUVmax was about 17.8. The maximum cross section and the SUVmax of the largest lymph node in the hepatic hilum were about 13.2 × 9.8 mm and 1.8. The lesion in right lobe of liver (white arrow): the maximum intensity projection PET image (A), transverse CT (B), and corresponding PET (C), and fusion images (D). The largest lymph node in the hepatic hilum (white arrow): transverse CT (E), and corresponding PET (F), and fusion images (G).

(Fig. 1E–G). No chaolangiectasis of liver inside and outside. PET/CT imaging suggested hepatic carcinoma in the right lobe of liver accompanied by lymph nodes metastasis in the hilus hepatis.

The patient underwent right hemihepatectomy and hilar lymph node dissection. Histological examination showed that the tumor in the right lobe of liver was separated by abundant fibrous tissues and existence of central sclerosis zone (Fig. 2A and B). Immunochemical staining of the mass in the right lobe of liver presented AFP and CK19 were negative, CD34, GPC3, GS, and HSP70 were positive, Ki-67 was 40% positive. The tumor was inclined to the sclerosing hepatocellular carcinoma. Interestingly, the histological image of lymph node in the hepatic hilum

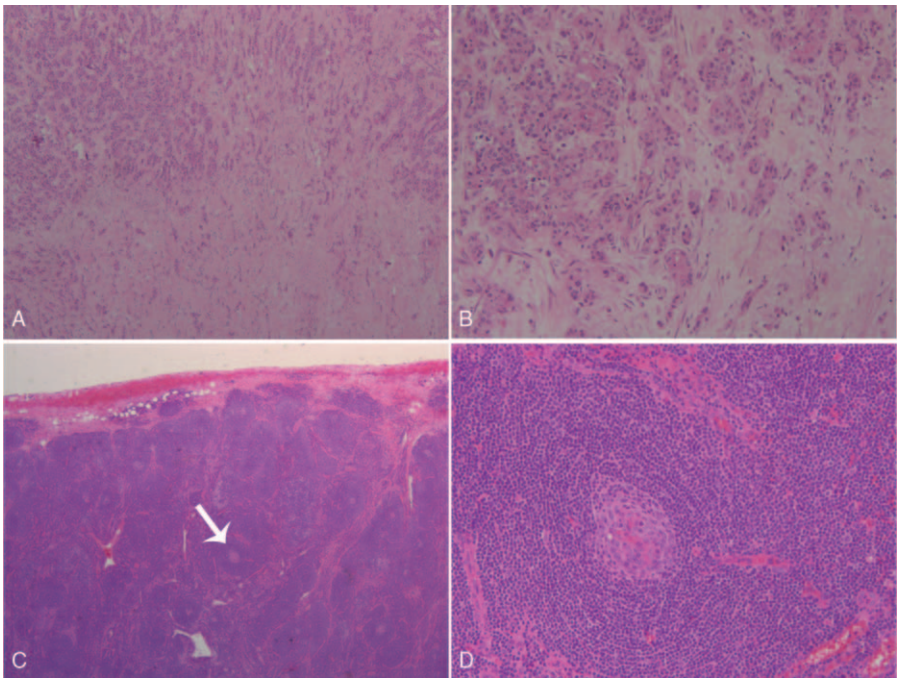

Figure 2. Histopathological images of the hepatic mass and the lymph node in the hilum of liver. Histological findings showed that the mass in the right lobe of liver prone to the sclerosing hepatocellular carcinoma (A, 40×; B, 200×). The histological image of lymph node in the hepatic hilum presented that lymph node structure existed, lymphoid follicles hyperplasia and germinal center atrophy, vascular proliferation were seen in the germinal center (white arrow). Hyaline-type Castleman disease in the hepatic hilum was suspected (C, 25×; D, 200×).

presented that lymph node structure existed, lymphoid follicles hyperplasia and germinal center atrophy, vascular proliferation was seen in the germinal center (Fig. 2C and D). The immunochemical examination of the mass in the hepatic hilum revealed CD3, CD5, CD10, CD20, CD79a, and Bcl-2 were positive, Ki-67 was 5% positive. Hyaline-type Castleman disease in the hepatic hilum was suspected.

3. Discussion

Castleman disease is a benign and rare lympholiferative disease, which was first reported by Castleman et al.[10] The common site of the disease is mediastinum, neck, abdomen, and axilla.[11] Castleman disease located in the hilum of liver has been reported in recent years.[1,3,5,12] However, these cases were unicentric Castleman disease which was a type of disease with a single lesion in the hilus hepatis. A case of hepatocellular carcinoma in the liver accompanied by Castleman disease has not been reported.

Castleman disease can be divided into 2 types according to pathologic characteristics: hyaline-vascular type and plasma-cell type. Hyaline-vascular type is the most common type in the Castleman disease characterized by hyperplasia of the hyaline-vascular follicles and capillary proliferation. The less common type is plasma-cell type characterized by plentiful plasma cell infiltration in the interfollicular tissue.[12] Clinically, Castleman disease is also classified into 2 types: unicentric type and multicentric type. The previous type is usually asymptomatic and often associated with a benign prognosis, but the later type is accompanied by systemic symptoms like fever, anemia, emaciation, generalized lymphadenopathy, hepatomegaly, and hepatosplenomegaly.[5]

Castleman disease often shows mild to moderate ^{18}F-FDG uptake and most patients often have no significant symptoms. In our case, the patient was no significant symptoms but for acratia and poor appetite, the serum level of tumor markers was no obvious abnormal increase, and the SUVmax of the largest lymph node in the hilum of liver was about 1.8. The lower uptake of ^{18}F-FDG may be affected by the tumor in the liver, and this guess needs more evidences to support in the following research. Therefore, it is hard to make a diagnosis between Castleman disease and lymph node metastasis when it accompanies by the hepatocellular carcinoma in the liver only depending on the imaging features by CT, MRI, or PET/CT. Anatomical and metabolic information in a single image can be obtained through one-step PET/CT examination. Compared to traditional imaging techniques, PET/CT has a more obvious advantage.[13] However, due to the particularity of Castleman disease, PET/CT has a limited specificity in the differential diagnosis between Castleman disease and lymphoma, lymph node tuberculosis, metastatic disease, sarcoidosis or multiple myeloma.[9,13–15] Castleman disease may be presented as lower uptake and not obvious in the ^{18}F-FDG PET/CT scanning, and hope our case provides a differential diagnosis for clinicians in the following similar cases.

Author contributions

Supervision: H. Shi.
Writing – original draft: Y. Hu.
Writing – review & editing: Y. Liu, H. Shi.

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