Incidental detection of rare mesenteric inflammatory pseudotumor by ^{18}F-FDG PET

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Abstract

A 60 years old asymptomatic male underwent fluorine-18 fluorodeoxyglucose positron emission tomography (^{18}F-FDG PET) for his medical check-up, and abnormal ^{18}F-FDG uptake was observed in the retroperitoneum. The maximum standardized uptake value (SUV_{max}) was 5.2. Based on CT, MRI and ^{18}F-FDG PET findings, the differential diagnosis included specific or non-specific inflammatory change, malignant lymphoma, trauma, gastrointestinal stromal tumor and soft-tissue sarcoma. Tumor resection was performed, and the histopathological finding was an inflammatory pseudotumor (IPT) originating at the mesentery in the retropetoneum. After two years and eight months from his initial operation, recurrent IPT was detected by ^{18}F-FDG PET for follow up, although he was asymptomatic. The IPT could be of traumatic origin since the patient suffered a severe abdominal trauma 6 months before. A mesenteric IPT is very rare, and to our knowledge, this is the first case report of ^{18}F-FDG PET detecting a mesenteric IPT. *In conclusion*, when abnormal high ^{18}F-FDG uptake is observed in the mesentery incidentally in clinical routine examination, IPT should be included as one of the differential diagnoses. ^{18}F-FDG may be useful in detecting local recurrence and follow-up after operation.

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Introduction

An inflammatory pseudotumor (IPT) is a rare benign lesion characterized by the nonneoplastic proliferation of inflammatory cells, predominantly plasma cells, and the presence of intermingling collagen fibers. This pseudotumor commonly occurs in the lungs and orbita, while a mesenteric IPT is very rare [1, 2]. Clinical symptoms are diverse according to the location of the lesion, such as tumor, fever, weight loss, and pain [1]. Patients commonly have symptoms and laboratory findings suggestive of inflammation; however, a definitive diagnosis is often difficult to make in the absence of clinical findings [3]. Recently, fluorine-18 fluorodeoxyglucose (^{18}F-FDG) has been reported to accumulate in IPT in the lung, spleen, liver, pancreas, colon, orbita, mediastinum, and central nervous system [4-10]. However, to our best of knowledge, ^{18}F-FDG has not been reported to accumulate in mesenteric IPT. We describe a rare case of asymtomatic mesenteric IPT incidentally identified by ^{18}F-FDG-PET.

Case report

A 60 years old asymptomatic male was receiving medical treatment for primary hypertension in the outpatient clinic of our institution. He underwent ^{18}F-FDG-PET for a medical check-up to screen for cancer. A dedicated full-ring PET scanner (Allegro: Philips Medical Systems, Inc., Cleveland, Ohio) consisting of a dedicated germanium oxyorthosilicate was used for data acquisition. Before ^{18}F-FDG injection, the patient had fasted for 4h to maintain serum glucose concentration below 120mg/dL. Intake of sugar-free liquids was permitted. Prior to the examination, the patient drank 500mL of tap water to accelerate renal ^{18}F-FDG elimination. His blood glucose levels were 88mg/dL. He was administered 252MBq (4.44MBq /kg) of ^{18}F-FDG via an antecubital vein. Whole-body PET imaging was performed at approximately 60min after ^{18}F-FDG injection. Transmission and emission images of the areas from the level of the auditory meatus to the mid-thigh were acquired (transmission images: 23sec, 10 bed positions; emission images: 2min and 30sec, 10 bed positions) with the patient in the supine position. Transmission scans were carried out to provide attenuation correction with a ^{137}Cs point source. After both transmission and emission images were obtained, the images were reconstructed using 3D-

RAMLA (3D-Row Action Maximum Likelihood Algorithm, Philips, Eindhoven, The Netherlands). The total examination time for whole-body PET images was approximately 30min. The ^{18}F-FDG PET scan showed abnormal uptake at the retroperitoneum in the abdomen. The maximum standardized uptake value (SUV max) was 5.2, and there was no abnormal ^{18}F-FDG uptake in other regions (Fig. 1). Abdominal contrast enhancement computed tomography (CT) showed tumor with peripheral contrast enhancement and high attenuation area of mesenteric adipose tissue (Fig. 2). Abdominal MRI showed the slightly high intensity tumor on T1-weighted image (T1WI), T2WI, and high intensity on short T1 inversion recovery (STIR). The diffusion-weighted MRI image showed decreased apparent deficient coefficient (ADC) value (Fig. 3). Laboratory findings revealed that interleukin 2 receptor (IL2R) was slightly increased, while other laboratory findings such as liver, kidney, and pancreas function were normal, and the resultant data of C-reactive protein (CRP), red blood cells, white blood cells, hemoglobin, electrolytes, and carcinoembryonal antigen (CEA) were within normal range. Non-specific inflammatory change was considered, because there was a high attenuation area of mesentery adipose tissue around the tumor on CT, and also malignant lymphoma, gastrointestinal stromal tumor (GIST), and soft-tissue sarcoma based on the radiological imagings and laboratory data. A tumor resection was performed, and the intra-operative finding revealed that the oval mesentery was thickened and the tumor lesion was located inside the mesentery. The pathological tumor size was 2.0X1.0cm, and the macroscopic finding of this tumor was solid and hard. Hematoxylin and eosin staining (HE staining) showed that the tumor was diffusely infiltrated by fusiform fibroblasts, macrophages, and lymphocytes (Fig. 4). The histopathological diagnosis was mesenteric IPT, with no malignant cells.

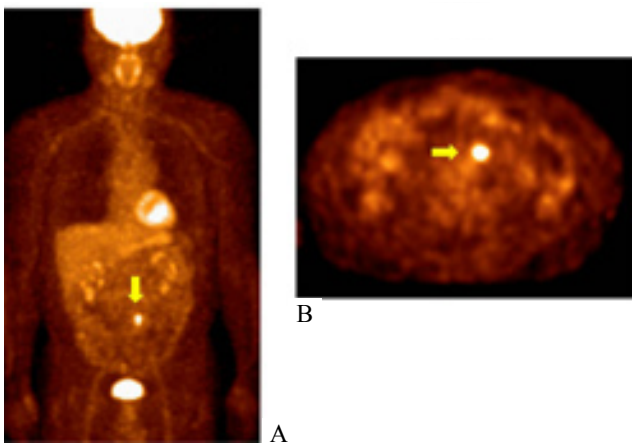

Figure 1. ^{18}F-FDG-PET before tumor resection: (A) Whole-body image: (B) Axial image**.** ^{18}F-FDG-PET showed abnormal uptake in the abdomen. The SUV_{max} was 5.2 (arrow). Other round shaped uptake lesions in the thorax and the abdomen observed on the whole-body image were considered as physiological ^{18}F-FDG uptakes of the liver, kidney, stomach and esophagus because there were no related abnormal findings on CT and endoscopy.

After the operation, the patient was followed up in the outpatient clinic at our institution. He underwent ^{18}F-FDG PET after two years and eight months from his initial operation, and abnormal ^{18}F-FDG uptake was detected in retroperitoneum (SUV_{max} 5.4) (Fig. 5). Abdominal contrast-en-

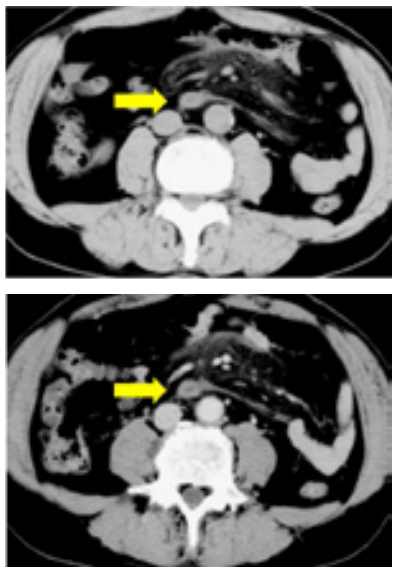

Figure 2. Abdominal CT before tumor resection. (A) Non-contrast CT showed the tumor lesion, 2cm in diameter, between the abdominal aorta and inferior vena cava. (B) Contrast-enhanced CT revealed the tumor with peripheral contrast enhancement (arrow).

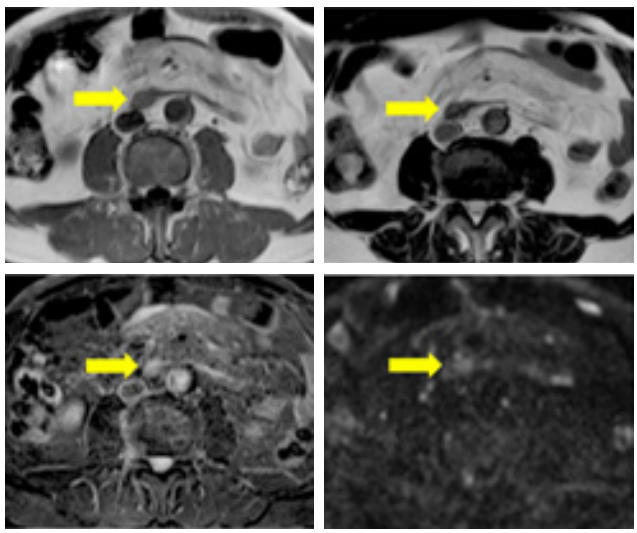

Figure 3. Abdominal MRI before tumor resection. MRI showed the tumor lesion with slightly high intensity on T1WI (A) and T2WI (B), and high intensity on short STIR (C). Diffusion-weighted MRI imaging identified a decline in diffusion capacity (arrow) (D).

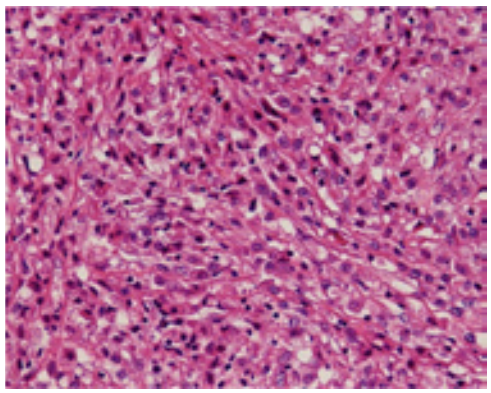

Figure 4. The histopathological findings after tumor resection. Hematoxylin and eosin staining (HE staining) revealed that the tumor was diffusely infiltrated by fusiform fibroblasts, macrophages, and lymphocytes. The histopathological diagnosis was mesenteric IPT, and it showed no malignant cells. Immnohistochemical staining showed no expressions of anaplastic lymphoma kinase (ALK), CD20, c-kit, IgG4, or Epstein-Barr virus (EBV).

hanced CT showed a tumor lesion in the mesentery, which was consistent with abnormal ^{18}F-FDG uptake. Recurrent IPT was diagnosed from CT and ^{18}F-FDG PET findings. This patient was asymptomatic had no treatment for the recurrent IPT and continued to be followed-up in the outpatient clinic. Follow-up CT was performed two years later, and the tumor lesion had disappeared completely.

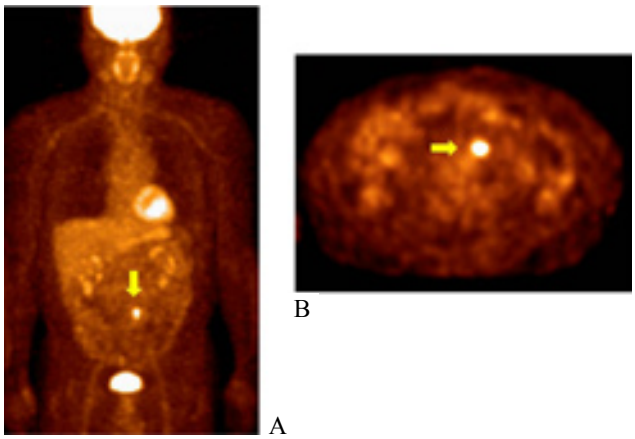

Figure 5. The recurrent tumor lesion of IPT detected by ^{18}F-FDG PET 2 years after tumor resection: (**A**) Whole-body image: (**B**) Axial image**.** ^{18}F-FDG-PET showed abnormal uptake in the abdomen. The SUV_{max} was 5.4 (arrow). Other round shape uptake areas seen on the whole-body image were considered physiological uptakes of the liver and kidney because there were no related abnormal findings on the CT image.

Discussion

An IPT is a benign rare tumor, and the clinical and imaging findings for an IPT are similar to those of a malignant tumor. The IPT are characterized by fibrous tissue infiltrated by inflammatory cells [11]. Ebstein virus infection (EBV) and IgG4 expression were negative [1, 6, 12]. This patient had no history of bacterial infection such as tuberculosis, and there were no histopathological findings of tuberculosis after operation. However, he had past history of thoracic and abdominal traumas due to fall from tree approximately six months before the ^{18}F-FDG-PET examination. We consider that the mesenteric IPT had been caused by that trauma.

There are many patients with mesenteric IPT among people in their twenties. The symptoms are abdominal pain, fever, and anemia [1]. The typical CT findings have been reported to be a heterogeneous low attenuation area, poor contrast enhancement, heterogenous contrast enhancement, and peripheral contrast enhancement [5].

Detection of IPT cases by ^{18}F-FDG PET shows high uptake as that in malignant tumors [4]. The mechanism of high ^{18}F-FDG uptake in IPT may be related to inflammatory cells in the pseudotumor [8]. As macrophages existed in the tumor on histopathological findings in this case, they may contribute to the mechanism of ^{18}F-FDG uptake in IPT.

Complete surgical resection for patients with IPT results in good prognosis. However, local recurrence rates of mesenteric or retroperitoneal IPT have been reported to be from 15% to 37% within one year after surgery, and strict follow-up is essential [13]. Our patient had local recurrence after two years from surgery, being still asymptomatic. His medical doctor discussed the management of recurrent IPT over with the patient. As he had no symptoms, he was followed-up without any therapeutic procedures. Others have reported that ^{18}F-FDG PET could detect IPT of the neck in multifocal sites [14] and IPT of the lung at the same time with a metastatic adrenal IPT lesion [15]. Furthermore, ^{18}F-FDG PET or PET/CT have been reported to be useful for evaluating the therapeutic effect of steroid therapy or radiation therapy for IPT in cases where surgical resection was incomplete [16, 17].

In conclusion, our case report suggests that abnormal high ^{18}F-FDG uptake in the mesentery may be due to IPT which in this case was due to trauma and recurred after dissection.

The authors declare that they have no conflicts of interest.

Bibliography

1. Coffin CM, Watterson J, Priest JR et al. Extrapulmonary inflammatory myofibroblastic tumor (inflammatory pseudotumor): a clinicopathologic and immunohistochemical study of 84 cases. *Am J Surg Pathol* 1995; 19: 859-72.
2. Narla LD, Newman B, Spottswood SS et al. Inflammatory pseudotumor. *Radiographics* 2003; 23: 719-29.
3. Fukuya T, Honda H, Matsumata T et al. Diagnosis of inflammatory pseudotumor of the liver: value of CT. *AJR Am J Roentgenol* 1994; 163: 1087-91.
4. Jeong JH, Cho IH, Kong EJ et al. ^{18}F-FDG PET/CT in inflammatory pseudotumor of the colon causing intussusception. *Ann Nucl Med* 2011; 25: 447-50.
5. Levy AD, Rimola J, Mehrotra AK, Sobin LH. From the archives of the AFIP: benign fibrous tumors and tumor like lesions of the mesentery: radiologic-pathologic correlation. *Radiographics* 2006; 26: 245-64.
6. Tsuboi H, Inokuma S, Setoguchi K et al. Inflammatory pseudotumors in multiple organs associated with elevated serum IgG4 level: recovery by only a small replacement dose of steroid. *Intern Med* 2008; 47: 1139-42.
7. Kawamura E, Habu D, Tsushima H et al. A case of hepatic inflammatory pseudotumor identified by FDG-PET. *Ann Nucl Med* 2006; 20: 321-3.
8. Sato M, Takasaka I, Okumura T et al. F-18 fluorodeoxyglucose accumulation in an inflammatory pseudotumor of the spleen. *Ann Nucl Med* 2007; 21: 521-4.
9. Huellner MW, Schwizer B, Burger I et al. Inflammtory pseudotumor of the lung with high FDG uptake. *Clin Nucl Med* 2010; 35: 722-3.
10. Yavuzer D, Dalbayrak S, Oz B et al. Intracranial inflammatory pseudotumor: case report and review of the literature. *Clin Neuropathol* 2010; 29: 151-5.
11. Horiuchi R, Uchida T, Kojima T, Shikata T. Inflammatory pseudotumor of the liver. Clinicopathologic study and review of the literature. *Cancer* 1990; 65: 1583-90.
12. Arber DA, Weiss LM, Chang KL. Detection of Epstein-Barr virus in inflammatory pseudotumor. *Semin Diagn Pathol* 1998; 15: 155-60.
13. Meis-Kindblom JM, Kjellstrom C, Kindblom LG. Inflammatory fibrosarcoma: update, reappraisal, and perspective on its place in the spectrum of inflammatory myofibroblastic tumors. *Semin Diagn Pathol* 1998; 15: 133-43.
14. Babar-Craig H, Gill H, Almeyda R et al. Inflammatory pseudotumour of the neck with multifocal sites on positron emission tomography scan imaging. *J Laryngol Otol* 2005; 119: 219-21.

15. Carolina C, Anile M, De Giacomo T, Venuta F. Bilateral simultaneous inflammatory myofibroblastic tumor of the lung with distant metastatic spread. *Interact Cardiovasc Thorac Surg* 2011; 13: 246-7.
16. Alongi F, Bolognesi A, Gajate AM et al. Inflammatory pseudotumor of mediastinum treated with tomotherapy and monitored with FDG-PET/CT: case report and literature review. *Tumori* 2010; 96: 322-6.
17. Obrzut SL, Halpern BS, Monchamp T et al. The role of 2-Deoxy-2[^{18}F] fluoro-D-glucose positron emission tomography/computed tomography in monitoring the immunosuppressive therapy response of inflammatory myofibroblastic tumor. *Mol Imaging Biol* 2004; 6: 126-30.