Isolated cholangiolitis revealed by ^{18}F-FDG-PET/CT in a patient with fever of unknown origin

Abstract

Cholangiolitis, inflammation of the cholangioles, is difficult to diagnose by conventional imaging modalities. We report a case of cholangiolitis revealed by fluorine-18 fluoro desoxyglucose positron emission tomography-computerized tomography (^{18}F-FDG-PET/CT) after about 9 months of recurrent fevers. A 20 years old girl with a history of recurrent fevers and repeated workups at different hospitals, which didn't diagnosed the source of fever, was admitted with a recent episode of fever. An ^{18}F-FDG-PET/CT was requested, which demonstrated focal hypermetabolic activity in the lateral segment of the left lobe of the liver. A liver biopsy showed inflammation of small biliary ducts consistent of cholangiolitis. Enterococcus casseliflavus was found on performed cultures. This represents the first case of cholangiolitis revealed by ^{18}F-FDG-PET/CT imaging.

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Introduction

The utility of metabolic fluorine-18 fluoro desoxyglucose positron emission tomography-computerized tomography (^{18}F-FDG-PET/CT) imaging in the evaluation of a variety of malignancies [1-4] has been well-accepted. It has been proposed that metabolic imaging is also valuable in the evaluation of infectious/inflammatory processes and searching the source of fever of unknown origin [5-8]. Cholangiolitis, which is inflammation of small terminal bile ductules, is poorly understood and difficult to diagnose. In this report, we described the ^{18}F-FDG-PET/CT finding of focal cholangiolitis in a 20 years old female with a 9 month history of fevers of unknown origin.

Case report

A 20 years old female with a 9 month history of intermittent fevers of unknown origin was admitted. She was diagnosed with congenital biliary atresia, status post Kasai procedure at 6 weeks of age, liver cirrhosis and portal hypertension. The patient had 9 admissions at different hospitals during the fever episodes. On her recent admission she was febrile with a temperature of 38.7°C. Routine laboratory studies showed an increased C reactive protein of 4.0mg/dL. In addition, there was mildly elevated bilirubin and decreased liver function, consistent with patient history of biliary atresia and liver cirrhosis. Repeated blood cultures showed no growth. Tests for HIV, Babesia and Plasmodium malaria, C. Trachomatis and N. Gonorrhoeae were all negative. In addition, respiratory virus polymerase chain reac-

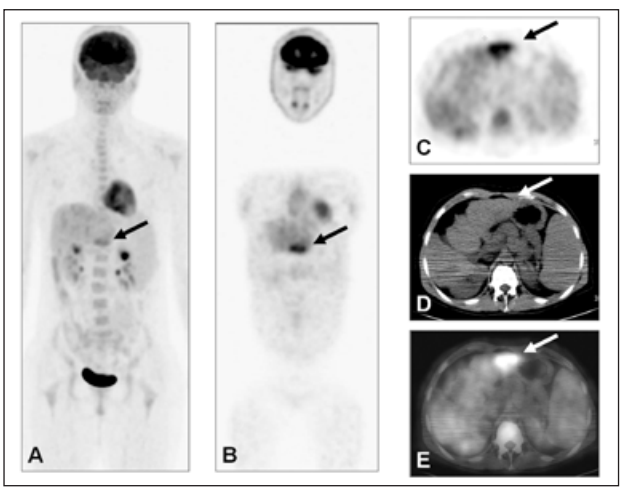

Figure A-E. Maximal projection image (**A**) and coronal image of the PET study (**B**) demonstrated an area of increased ^{18}F-FDG activity in the upper abdomen (arrows). On transaxial image (**C**), this abnormally increased activity (arrow) corresponded to the lateral part of the left lobe of the liver on CT (**D**) and fusion (**E**) images.

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tion (PCR) panel was negative for Adenovirus, Respiratory syncytial virus, Rhinovirus, Metapneumovirus, Influenza and Parainfluenza viruses.

The anatomical imaging examinations, including abdominal contrast-enhanced CT and ultrasound, demonstrated cirrhosis, portal hypertension and splenomegaly, which were expected findings in this potential liver transplant candidate. No abscess or potential sources of fever were detected.

An ^{18}F-FDG-PET/CT scan was subsequently performed to further evaluate this patient. The PET images demonstrated focally increased ^{18}F-FDG uptake with a maximum standardized uptake value of 4.3 in the anterior abdomen (arrows, Fig. A-C), located in the lateral segment of the left lobe of the liver on the corresponding CT image (arrows, Fig. D-E) which showed cirrhotic changes but no other abnormalities typical of focal inflammation or malignancy.

In order to determine the nature of the abnormal ^{18}F-FDG activity in the left lobe of the liver, a percutaneous liver biopsy with biliary duct aspiration was performed. Histopathologic examination of the biopsied liver specimen demonstrated ductular proliferative reaction with inflammation associated with mild cholestasis and portal fibrosis with focal bridging. The findings were consistent with cholangiolitis. The aspirated bile tested negative for cytomegalovirus (CMV) and Epstein-Barr virus (EBV) by real-time PCR. However, in the subsequent tissue culture of the biopsied liver grew Enterococcus Casseliflavus. Proper antibiotics treatment was initiated. The patient's symptoms promptly improved and she was discharged.

Discussion

The true incidence of cholangiolitis remains uncertain and chlangiolitis is often under-diagnosed, mainly due to lack of effective non-invasive diagnostic methods. Its etiology can vary from infectious, autoimmune or allergic, to drug toxicity and cholestasis. It has been commonly described in children with congenital liver fibrosis and reported in as many as 80% of cases of hepatic necrosis in children and infants [9, 10]. Isolated cholangiolitis can remain undiagnosed for long periods of time, despite repeated admissions and extensive investigations [9, 10]. The findings from our case indicate that ^{18}F-FDG-PET/CT has a potential role to play in diagnosis.

Increased ^{18}F-FDG activity focally in the liver caused by different malignant [11-16] or benign [17-20] processes has been reported. Increased ^{18}F-FDG activity in the biliary system due to different etiologies has also been reported previously [11, 12, 21-23]. However, to the best of our knowledge, findings of cholangiolitis revealed by ^{18}F-FDG-PET/CT have not been previously reported.

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