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Case Report

Dual-phase FDG-PET Imaging Shows Suspected Malignancy That Histological Examination Later Confirmed as Sclerosing Mediastinitis: Report of a Case

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Sclerosing mediastinitis is a rare, benign disorder that is often indistinguishable from malignancy by conventional imaging techniques. The value of fluorodeoxyglucose positron emission tomography (FDG-PET) imaging in the diagnosis of this disorder has not been elucidated. Recently, a few studies have reported the use of dual-phase FDG-PET imaging in the diagnosis of malignancies. The dual phase contains early- and late- phase images. The maximum standard uptake value (SUVmax) of late phase images of malignant lesions tends to be higher than those of early phase images. The present case showed that early-phase SUVmax was 5.93, and late phase, 8.92. We strongly suspected malignancy from the results of this new imaging technique, though the histological examination of the surgical samples provided the definitive diagnosis of sclerosing mediastinitis, Flieder stage II. This report describes the uncommon use of dual-phase FDG-PET computed tomography in the preliminary diagnosis of sclerosing mediastinitis. It is thought that current imaging studies are insufficient for the diagnosis of sclerosing mediastinitis, and the rarity of this disorder may prevent the development of imaging techniques. Histological confirmation is still essential for the definitive diagnosis of this disorder.

Key words: sclerosing mediastinitis, fibrosing mediastinitis, fluorodeoxyglucose positron emission tomography, mediastinal tumor

Introduction

Sclerosing mediastinitis, also known as fibrosing mediastinitis or mediastinal fibrosis, is a rare benign disorder characterized by the proliferation of dense fibrous tissue in the mediastinum.[1, 2] Its compressive progression or invasion of mediastinal structures mimics malignant tumors.[3] Positron emission tomography (PET) imaging with 2-[^{18}F]-fluoro-2-deoxy-D-glucose (^{18}F-FDG) produces varied results when used to examine sclerosing mediastinitis.[4–6] We present a case of sclerosing mediastinitis in the anterior mediastinum that was strongly suspected of being a malignant disorder based on the findings of dual-phase ^{18}F-FDG-PET computed tomography (CT) imaging.

Case Report

Complaining of severe abdominal pain, a 36-year-man with an unremarkable medical history was admitted to another hospital. Chest and abdominal CT scan revealed a mediastinal abnormality. Even though his abdominal

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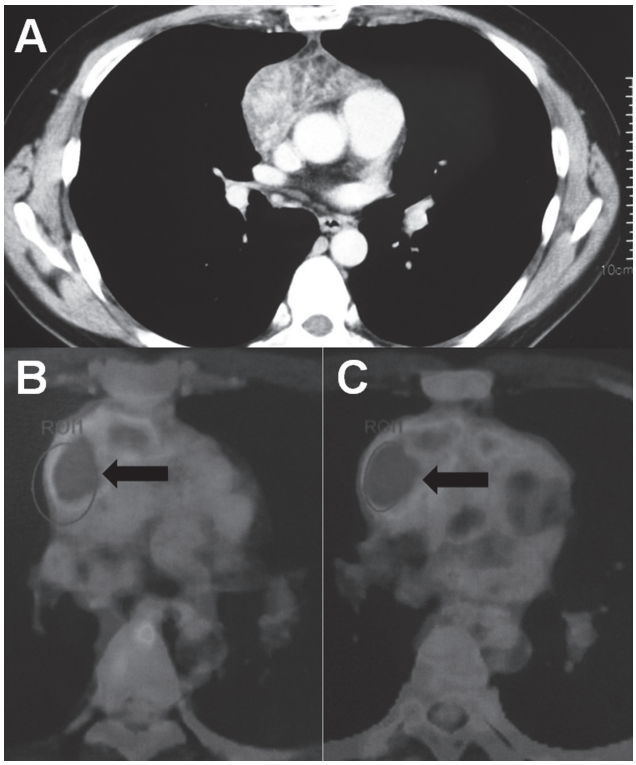

Fig. 1 (**A**) Chest computed tomographic (CT) scan showing anterior mediastinal swelling. (**B**, **C**) Dual phase ^{18}F-FDG-PET CT images. (**B**) Image of the early phase (60 min) shows the positive up-take (arrow). SUVmax is 5.93. (**C**) Image of the late phase (120 min) SUVmax is 8.92.

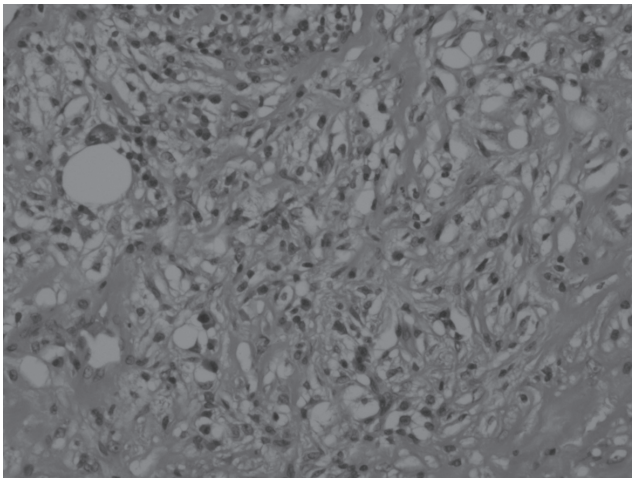

Fig. 2 Histological evaluation of the mediastinal disorder. The lesion contains fibrosis connective tissue in which aggregates of lymphocytes and plasma cells are found. This is consistent with stage II sclerosing mediastinitis. (Hematoxylin-eosin stain, 400 ×)

pain soon passed, the patient came to Tsuchiura Kyodo General Hospital for a more detailed examination of this mediastinal abnormality. Physical examination was unremarkable. The white blood cell count was 14690/mm^3 with 78% neutrophils. The level of C-reactive protein had increased to 5.2 mg/dl. Chest CT demonstrated an abnormal lesion, 9 × 3 × 11 cm in size, located in the anterior mediastinum (**Fig. 1A**). Magnetic resonate image (MRI) showed that it consisted of solid and fat areas. The lesion seemed to be a mediastinal swelling, such as thymic hyperplasia, rather than a malignant disorder, such as thymic epithelial tumor or germ cell tumor. The possibility of malignancy could not be entirely ruled out from the CT and MRI findings. Dual phase ^{18}F-FDG-PET/CT was used to help distinguish between the benign and malignant disorders. The dual phase contains early and late phase images. The early phase image was performed 60 minutes after the injection of ^{18}F-FDG, and the late phase image was obtained 120 minutes after the injection. The maximum standard uptake value (SUVmax) of the lesion was calculated with the dual phase. The SUVmax of the early phase was 5.93, and late phase was 8.92 (**Fig. 1B** and **1C**). The higher SUVmax of the late phase led us to strongly suspect a malignant mediastinal disorder. The presence of lung between the chest wall and mediastinal disorder made it impossible to carry out a percutaneous needle biopsy, thus, histological examination was necessary for making the diagnosis and creating a strategy for treatment. We resected this lesion through a median sternotomy, in the same way as an extended total thymectomy is carried out, for a precise histological diagnosis and as a surgical treatment, at two months after the first consultation. The anterior mediastinal tissues, swelling diffusely and tightly, could be thoroughly resected macroscopically. Histological examination showed the lesion mainly contained strong fibrosis connective tissue, in which aggregates of lymphocytes and plasma cells were found. Immunohistochemical stains and molecular biological examination ruled out malignant diseases such as malignant lymphoma, thymic epithelial tumors, and others. Finally, the diagnosis of sclerosing mediastinitis was made and this was classified as stage II according to the Flieder classification (**Fig. 2**).[7] Twelve months post-surgery, the patient is doing well with no recurrent symptoms of the sclerosing mediastinitis.

Discussion

Sclerosing mediastinitis is a rare, benign disorder in which dense fibrous tissue proliferates in the mediastinum.

While many such cases in the United States have been linked to *Histoplasma capsulatum* infection, many other causes can bring about this disorder, including fungal infection, tuberculosis, sarcoidosis, traumatic hemorrhage, drugs, malignancies, autoimmune disease, and idiopathic disorders.[1, 2] Since our findings were negative for histoplasma antibody in the serum and the patient had had no previous diseases, the pathogenesis of our case remains unclear. Diagnosis for sclerosing mediastinitis requires clinical suspension and multiple imaging studies, including CT or MRI, but the findings are often similar to those of malignant disorders, when using conventional imaging techniques.[1, 3] While some case reports have described FDG-PET findings on sclerosing mediastinitis, the value of these remains largely unclarified. Some reports have shown FDG-PET positive sclerosing mediastinitis,[4] and others negative.[6] Recently, a few studies have reported the use of dual-phase ^{18}F-FDG-PET imaging for diagnosing malignant disorders. It has been deduced that the difference in the time course of ^{18}F-FDG uptake could be used to improve the ability of PET to distinguish benign lesions and malignant ones.[8] Briefly, the SUVmax of the late phase images tends to be higher than the early phase images in malignant lesions. No one has reported applying this new technique to sclerosing mediastinitis. The higher SUVmax value in the late phase in this case suggests that the dual-phase ^{18}F-FDG-PET also failed to distinguish sclerosing mediastinitis from malignant diseases. It is thought that no current imaging studies can be used in the definitive diagnosis of sclerosing mediastinitis, and the rarity of this disorder may prevent the development of imaging diagnoses for it. In the end, histological confirmation is still essential in the definitive diagnosis of sclerosing mediastinitis. ^{18}F-FDG-PET is useful for determining the extent of sclerosing mediastinitis[5] rather than for differentiating malignancy.

There is no standard treatment or strategy for this disorder, and its treatment is controversial. Surgery, tamoxifen, and steroids have all been reported as efficacious strategies.[1, 3, 9] The indication for surgery, in most cases, is made in order to establish the diagnosis.[9] Some patients have had severe compression syndromes brought on by this disorder[3]; and since the patient described here might also develop compression syndrome as the disorder progresses, we thought that a total resection through mediansternotomy would be completely effective for the patient.

In conclusion, this is the first report of the use of dual-phase ^{18}F-FDG-PET/CT imaging for sclerosing mediastinitis. Sclerosing mediastinitis could not be distinguished from malignancy with this technique. Histological confirmation is still essential for precise diagnosis of this disorder.

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