Clinical Case Report

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Unusual synchronous liver and brain abscesses infected by rare *Aerococcus viridians* in a patient with pulmonary arteriovenous malformations on FDG PET/CT

A case report and literature review

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Abstract

Rationale: Pulmonary arteriovenous malformations (PAVMs) complicated with multiple organ abscesses is an uncommon manifestation. Because of the low incidence of the disease, F-18 fluorodeoxyglucose positron emission tomography with computed tomography (^{18}F-FDG PET/CT) imaging studies for PAVMs complicated with multiple organ abscesses are scarce.

Patient concerns: We report a case of a 54-year-old man presenting with PAVMs complicated with synchronous multiple organ abscesses founded by ^{18}F-FDG PET/CT. ^{18}F-FDG PET/CT revealed tortuous stripes and mass opacities with no significant FDG uptake in the left upper lung lobe. However, hypermetabolic lesions located in the anterior inferior segment of right hepatic lobe [with maximum standardized uptake value (SUVmax) of 10.7], and in the right basal ganglia with SUVmax of 14.1 were found by ^{18}F-FDG PET/CT.

Diagnoses: A diagnosis of synchronous liver and brain abscesses infected by rare *Aerococcus viridans* was determined by tissue culture.

Interventions: Vancomycin was provided intravenously, and oral linezolidate tablets were prescribed for anti-inflammatory treatment for 1 month. Liver and head magnetic resonance imaging was performed during the follow-up.

Outcomes: The lesion in the right basal ganglia was reduced, and the lesion in the right liver had disappeared, indicating the lesions were abscesses.

Lessons: The present case indicated that the possibility of abscesses should be considered with patients with PAVMs, and whole-body ^{18}F-FDG PET/CT is suggested to identify possible accompanying abscesses in multiple organs for PAVMs patients.

Abbreviations: ^{18}F-FDG = F-18 fluorodeoxyglucose, HHT = hereditary hemorrhagic telangiectasia, MR = magnetic resonance, PAVM = pulmonary arteriovenous malformation, PET/CT = positron emission tomography/computed tomography, SUVmax = the maximum standardized uptake value, T2WI = T2-weighted imaging.

Keywords: abscesses, *Aerococcus viridans*, liver, positron emission tomography/computed tomography, pulmonary arteriovenous malformations

1. Introduction

Pulmonary arteriovenous malformations (PAVMs) are rare vascular diseases of the lung where an abnormal communication exists between the pulmonary artery and vein. A direct path between the lung circulation and systemic circulation results in a right-to-left shunt. Clinical manifestation of PAVMs may be asymptomatic or may show symptoms such as cough, chest pain, palpitation, epistaxis, hemoptysis, and difficulty breathing. In severe cases, complications such as hypoxemia, thrombosis, cerebral vascular accident, brain abscesses, and bronchial artery rupture could occur. Some patients have accompanying hereditary hemorrhagic telangiectasia (HHT) or Osler–Weber–Rendu syndrome. Here, we report a case of rare PAVMs complicated with synchronous brain and liver abscesses examined by ^{18}F-fluorodeoxyglucose positron emission tomography/computed tomography (^{18}F-FDG PET/CT), along with a literature review.

2. Case report

The patient was a 54-year-old man seeking medical attention due to fever and cough that had lasted for 1 week. His body

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This study protocol was approved by the Institutional Review Board of Zhejiang University. Written informed consent was obtained from the patient.

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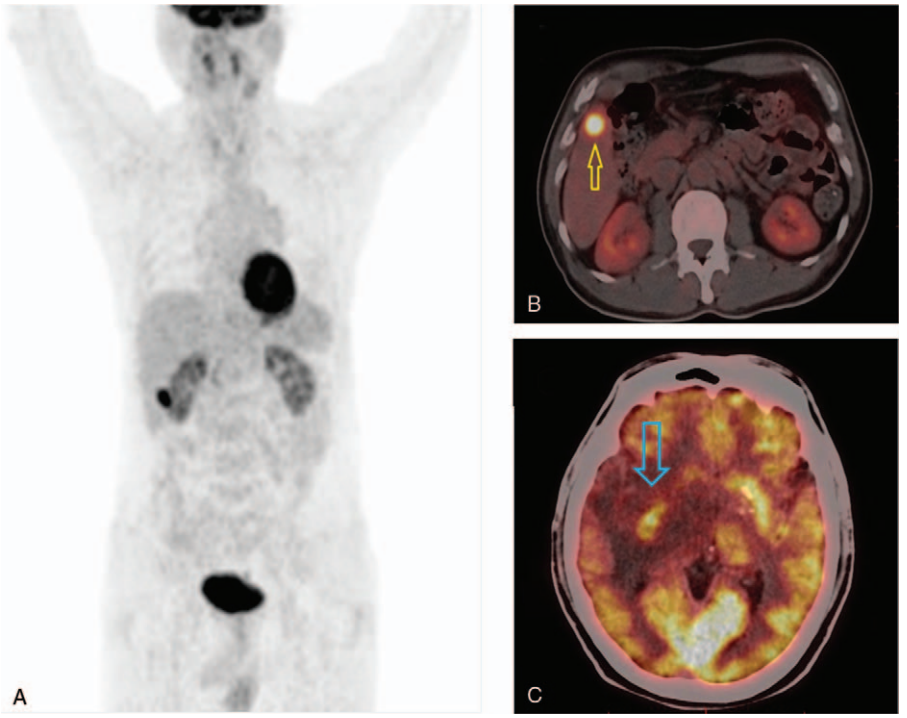

Figure 1. F-18 fluorodeoxyglucose positron emission tomography with computed tomography (^{18}F-FDG PET/CT), positron emission tomography (PET) body maximum intensity projection (MIP) image (A), and abdominal PET/CT (B) revealed a focal FDG metabolic nodule (yellow hollow arrow). Head PET/CT (C) identified a basal ganglia heterogeneous nodule with increased FDG uptake, and a zone of edema was observed with the centreline shifted slightly toward the left (arrow).

temperature was 38°C at the highest. Coughing with sputum was an apparent symptom and the sputum color was white and moderate in quantity. There were no apparent symptoms such as headache, chest pain, palpitation, chest tightness, shortness of breath, and hemoptysis. A plain lung CT scan suggested a mass in the left lung, and head magnetic resonance (MR) imaging suggested a mass in the right basal ganglia. The tentative clinical diagnosis was lung malignancy complicated with brain metastasis. The patient was admitted to our hospital ward for further diagnosis and treatment.

A physical examination revealed that there were no apparent discolorations on the skin, sclera, or oral mucosa. The superficial lymph nodes were not enlarged. The patient's breath sounds in the lungs were heavy, but there were no apparent moist and dry rales. The heart maintained a stable rhythm and without any apparent murmur. Hematological and biochemical laboratory test results were as follows: blood gas analysis for oxygen partial pressure (PaO_2) was 63.1 mm Hg, the erythrocyte sedimentation rate was increased (14 mm/h), and C-reactive protein was 9.0 mg/L; negative results were obtained for all other hematological and biochemical tests. To further confirm the diagnosis, an ^{18}F-FDG PET/CT examination was performed. The ^{18}F-FDG PET/CT revealed a 47 × 35 mm irregular nodule, with tortuous and mass-occupying lesions, exhibiting no increased F-18 FDG uptake in the left upper lung lobe. An isolated low-density nodule was found on the right anterior of the liver; the size of the lesion was approximately 23 × 21 mm; the density was fairly even, but the boundaries were not clear. FDG uptake increased abnormally with the maximum standardized uptake value (SUVmax) of 10.7. We also found a soft-tissue density mass accompanied by a zone of edema in the right basal ganglia and significantly uneven ^{18}F-FDG uptake with the SUVmax of 14.1. The right lateral ventricle was pressurized (Fig. 1).

Lung enhanced CT and angiography was subsequently performed, and proved upper left PAVMs. The upper and lower pulmonary arteries communicated with the pulmonary vein. The left subphrenic artery participated in supplying blood and connected with the left gastric artery in the abdominal cavity. Scattered small calcifications could be seen on the internal wall without apparent filling defects (Fig. 2). The lesion on the right anterior of the liver showed mix signal intension on T2-weighted imaging (T2WI) on liver MR (Fig. 3C). Head MR imaging suggested abnormal stripes in the right basal ganglia. T1-weighted imaging showed low intensity, whereas T2WI and diffusion-weighted imaging showed high intensity. Ring and nodular enhancements were observed (Fig. 3A). Therefore, the possibility of malignant hepatic tumour complicated with brain metastasis was highly suspected.

Ultrasound-guided liver lesion biopsy was subsequently performed. Histologic findings of the specimen revealed inflammatory cell infiltration, yet no evidence of malignancy was noted (Fig. 4). It is interesting that a high amount of *Aerococcus viridans* was found in the tissue culture. Drug sensitivity testing suggested that the bacteria were vancomycin sensitive. So vancomycin was provided intravenously, and oral linezolidate tablets were prescribed for anti-inflammatory treatment for 1 month. Liver and head MR imaging was performed during the follow-up. The lesion in the right basal ganglia was reduced, and the lesion in the right liver had disappeared (Fig. 3B, D), indicating the lesions were abscesses.

3. Discussion

Approximately 5% to 10% of PAVMs are complicated with brain abscesses, often located at the basal ganglia. Such

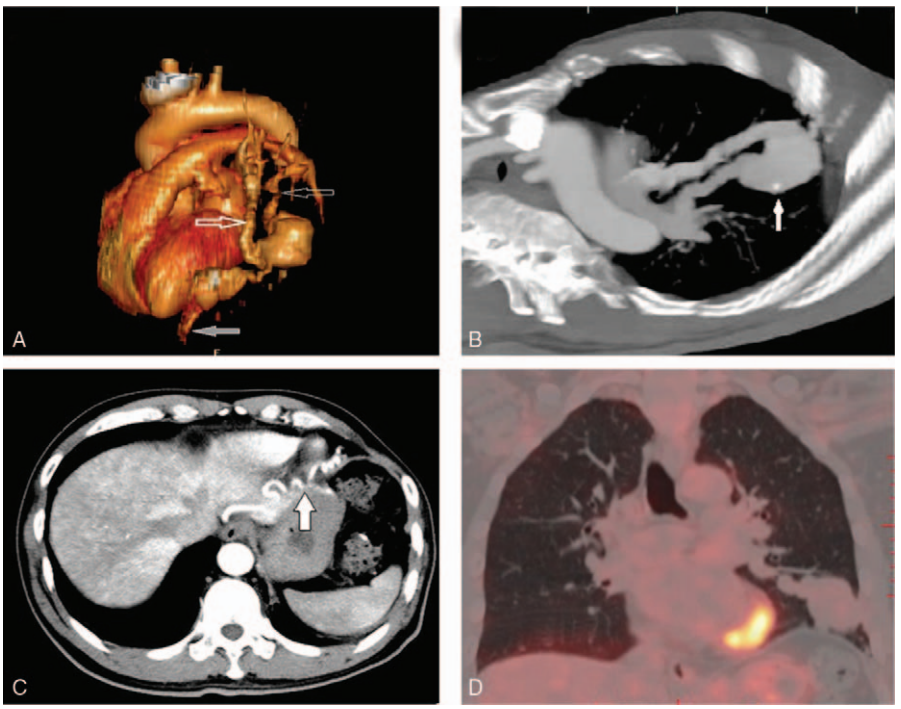

Figure 2. Chest CT vessel reconstruction showed abnormal branch vessel of the pulmonary arteriovenous malformations (PAVMs; grey solid arrow), vein (white hollow narrow arrow), and artery (white hollow thick arrow) (A). Calcification was presents in the lesion (B). Lung-enhanced CT scan identified an abnormal tortuous vessel at the left diaphragm that connects the left PAVM and the left gastric artery and forms a systemic pulmonary artery anastomosis (C). F-18 fluorodeoxyglucose positron emission tomography with computed tomography (^{18}F-FDG PET/CT) revealed enlarged mass with no significant F-18 FDG uptake (maximum standard uptake value of 1.4) in the left upper lung lobe (D).

conditions are related to the right-to-left shunt resulting from arteriovenous malformations.[1,2] Press and Ramsey[3] reported that for patients with PAVMs accompanied by HHT, 29/31 (94%) of the cases were complicated by brain abscess or meningitis. However, complications with abscesses in other organs and tissues are rarely previously reported. Haarmann *et al*[4] reported a case of PAVMs complicated by subcutaneous temporal and brain abscesses. The patient had HHT, and telangiectasia was observed on the tongue, lips, and oral mucosa. To the best of our knowledge, FDG PET/CT studies of PAVMs that are complicated by multiple organs and tissues abscesses are rarely reported.

Possible reasons of synchronous liver and brain abscesses infected by rare *A viridians* in a patient with PAVMs may be as follows: Systemic-pulmonary artery anastomosis with abnormal septic emboli—There was an abnormal branch in the chest-abdominal region. Enhanced CT revealed an abnormal tortuous vessel at the bottom of the PAVM that was connected to the left gastric artery in the abdominal cavity. Yet, this vessel could also have been an abnormally thickened left subphrenic artery (Fig. 2). A systematic-pulmonary artery branch was formed. Normally, the diameter of the subphrenic artery is 1.3 ± 0.1 mm. The septic embolus of a PAVM is mixed with abdominal arterial blood under pulmonary arterial pressure, which can be carried to the liver, where it can form liver abscesses.[5] Cardiogenic liver disease—Pulmonary venous blood flowed into the abdominal artery blood via the branch, which resulted in a reduction in the oxygen supply in the pulmonary artery. In addition, substances such as CO_2 and 5-hydroxytryptamine can flow to the hepatic artery through the venous system.[6] The result is a reduction in the capacity to regulate hepatic blood and hepatocellular ischemia or hepatocellular damage. Therefore, the capacity of the liver to eliminate metabolic products and the immunity are reduced, further facilitating abscess in the liver. The possibility of patients with PAVMs accompanied by HHT—It is reported that approximately 47% to 88% of patients with PAVMs have accompanying HHT.[7] As reported in the literature when comparing patients with HHT to those without HHT, patients with HHT are more likely to have infectious diseases, such as meningitis and liver abscesses; however, the mechanisms are still unclear.[8] The early manifestation of liver HHT is widening of the venules in the liver, which can be seen under a microscope. This continuous vessel damage can result in multiple arteriovenous fistulas. The pathological manifestation is focal blood sinus expansion and internal fistulas. Liver blood flow increases because of the fistulas so that it is possible to identify hepatic artery expansion in early stages. In enhanced spiral CT images of the early arterial phase, small vascular malformations typically appear as dotted enhancement in the liver.[9,10] In our case reported here, the enhanced CT image of the liver in the arterial phase had scattered uneven patchy enhanced lesions. Small liver vascular abnormalities or arteriovenous fistulas were possible. The diverging path due to arteriovenous fistulas reduced the blood flow to the liver and the biliary system. The liver's blood supply was then changed, which caused damage to the liver and bile duct cells. To a certain degree, this damage also facilitated the occurrence of liver abscess. Normal results in liver indexes, such as bilirubin, transaminase, alkaline phosphatase, and glutamyl transpeptidase, could be due to compensatory effects. Biological factors—Liver abscesses caused by *A viridans* infections are rarely seen in the literature. *A viridans* is a type of conditionally pathogenic bacteria, and invasive infections are rarely seen.

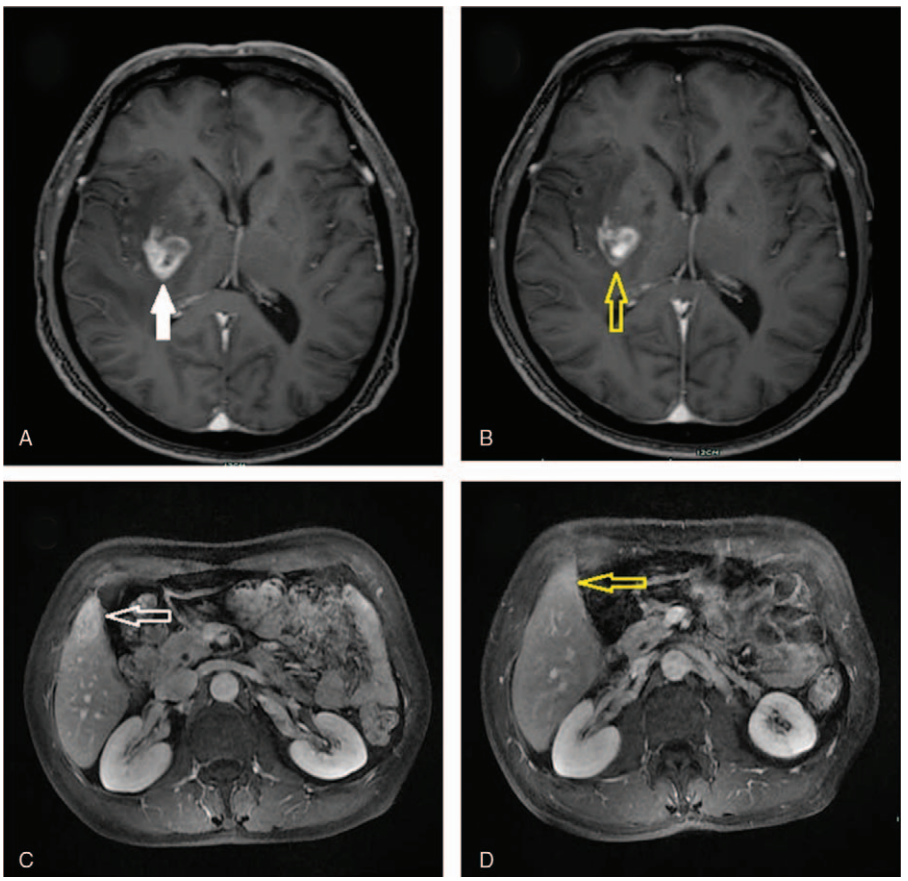

Figure 3. Head enhanced magnetic resonance (MR) image before treatment revealed an apparent heterogeneously enhanced nodule (solid arrow) in the right basal ganglia (A), and decreased in size of the enhanced nodule (hollow arrow) after 1 month of anti-inflammatory treatment (B). Liver-enhanced MR image before treatment revealed the heterogeneous enhanced nodule (arrow) on the right anterior of the liver during the venous phase (C), and disappeared lesion after anti-inflammatory treatment (D).

However, patients with conditions such as low immunity, open wounds, and intravenous catheters are easily infected. The infection often results in endocarditis, suppurative arthritis, bacteraemia, meningitis, and other conditions.[11,12] Zhou and Peng [13] revealed that infection sites of *A viridans* in clinical cases are commonly seen in biliary tract infections (10.5%). For the patient in our case reported here, the arteriovenous malformation resulted in a longer time for the right-to-left shunt. The body was in a state of mild hypoxia (PaO_2 was 63.1 mm Hg). This condition could have resulted in reduced immunity, which could have caused the infection. Arteriovenous malformation—Many reports in the literature have discussed the correlation between potential odontogenic bacteraemia and abscess formation. Because of the direct flow of blood through the PAVMs, the engulfing function of the reticuloendothelial cells of the pulmonary capillary bed is partially lost. The capacity to eliminate bacteria is then lowered, which could be beneficial for bacterial replication and growth, which could result in an abscess.[14,15]

In our case reported here, FDG PET/CT in combination with enhanced CT was implemented for the diagnosis of PAVMs. In addition, a focal high metabolic lesion was found in the right lobe of liver along with an intracranial basal ganglia metabolic lesion. Multiple diseases needed to be differentiated, especially intrahepatic cholangiocarcinoma and isolated liver abscesses. When lesions are small and without apparent necrosis, FDG uptake is abnormally increased. Therefore, diagnosis requires comprehensive clinical and laboratory tests and enhanced MR imaging. In recent years, due to the misuse of antibiotics, the clinical and imaging features of liver abscesses are atypical; thus, issues in

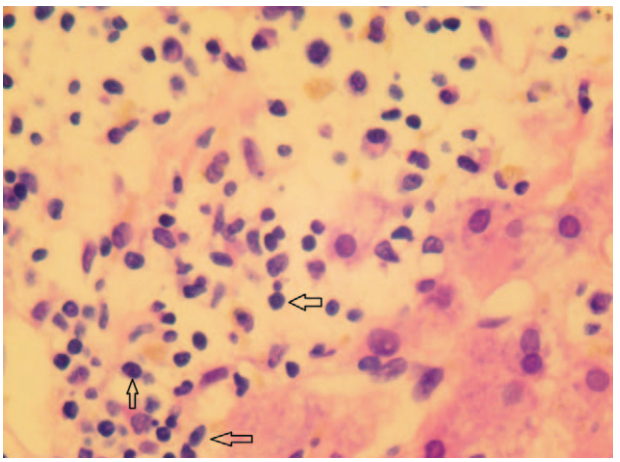

Figure 4. Liver biopsy (HE × 400): A small amount of liver tissue had localized myofibroblast hyperplasia. Inflammatory cell (black arrow) infiltration was observed. HE = hematoxylin-eosin staining.

differentiating abscesses from malignant liver tumours or cases of misdiagnosis have increased.[16]

For treatment, intravascular embolization and surgical removal can be performed for PAVMs. Surgical removal is the primary choice for single lesions, whereas intravascular embolization is better suited for multiple lesions.[17,18] For patients with confirmed abscesses, anti-infectious treatment should be provided based upon drug sensitivity testing. However, it should be noted that the size, degree, and symptoms of PAVMs cannot be the bases for predicting the occurrence of complications, such as abscesses and cerebral vascular accidents; therefore, prevention is very important.[19,20] Tobacco use should be prohibited; blood pressure, blood sugar, and cholesterol should be controlled; and atrial fibrillation should be prevented.[21] There are also reports regarding the need for preventive antibiotic use and improvement of oral cavity health to reduce the risk of bacteraemia and abscesses.[22,23]

4. Conclusion

The present case indicated that the possibility of abscesses should be considered with PAVMs patients; whole-body ^{18}F-FDG PET/CT is feasible modality to identify possible accompanying abscesses in multiple organs for PAVMs patients.

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