density in both lungs, which was confirmed on chest computed tomography (CT) (Fig. 1C). An incidental finding was bilateral pulmonary thromboembolism (Fig. 1D). PJP was suspected, so fiberoptic bronchoscopy was performed which confirmed PJP in bronchoalveolar lavage. The patient responded favorably, both clinically and radiologically, to anticoagulants and antibiotic treatment with trimethoprim–sulfamethoxazole.

PJP is a serious, opportunistic infection, commonly seen in patients with advanced HIV infection, but it can also affect patients who are immune deficient for other reasons (high corticosteroid doses or immunosuppressants, anticancer treatments, etc.). In recent years, the incidence of PJP has risen in non-HIV patients. Moreover, in patients without HIV, PJP is more aggressive and generally has a higher mortality rate than in patients with HIV infection.[4] Temozolomide is a relatively new alkylating anticancer drug used in the treatment of high-grade glial tumors. One of its effects is to induce lymphocytopenia and T cell dysfunction, predisposing patients to developing PJP. This complication is particularly common in patients receiving concomitant corticosteroids and radiation therapy (2 standard treatments in patients with central nervous system tumors), so the prophylactic administration of trimethoprim–sulfamethoxazole is usually recommended to prevent its appearance.[2] Thromboembolic disease is also relatively common in patients with glioblastoma (up to 33% of GM patients develop deep vein thrombosis and/or pulmonary embolism, according to a recent study), particularly in the first month after neurosurgery and during chemotherapy.[5] Few references are available in the literature that describe PJP in patients receiving temozolomide and to our knowledge none has described concomitancy with pulmonary thromboembolism.

We believe that knowledge of this specific infectious respiratory complication in GM patients receiving temozolomide is important for early detection and optimal management.

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Autopsy Case of Pulmonary Artery Sarcoma Forming Aneurysm Without FDG Uptake☆

Autopsia de un sarcoma de arteria pulmonar, que formaba un aneurisma sin captación de FDG

To the Editor:

Pulmonary artery (PA) sarcoma is a rare tumor,[1] with clinical symptoms and radiological findings that often resemble pulmonary emboli.[2] Main PA aneurysm is an uncommon presentation of PA sarcoma. To the best of our knowledge, only 2 cases have been reported,[3,4] neither of which was evaluated with 2-deoxy-2-[18F]fluoro-D-glucose (FDG) positron emission tomography (PET)/computed tomography (CT). We report a case of main PA aneurysm due to PA sarcoma without FDG uptake. This is the first autopsy case report of PA sarcoma with main PA aneurysm.

A 38 year-old-man was referred to our hospital for right main PA aneurysm and well-defined multiple nodules in both lungs on chest CT. PET/CT showed FDG uptake in multiple lung nodules, but not in PA aneurysm (Fig. 1A and B). Transthoracic echocardiography and transesophageal echocardiography revealed PA aneurysm compressing left atrium. PA aneurysm due to PA sarcoma with multiple lung metastases was suspected, and the patient underwent thoracoscopic biopsy and pericardial fenestration. PA leiomyosarcoma was diagnosed on the basis of microscopic findings. Surgery was ruled out due to the prognosis and the invasiveness of surgical management. The patient received chemotherapy with pazopanib, which was stopped due to an allergic reaction with rash and fever. He was scheduled for second line chemotherapy, but died suddenly 2 months after the diagnosis. An autopsy was performed with the consent of his family about 26 h after death.

The autopsy revealed main PA leiomyosarcoma with cystic degeneration (Fig. 1C) and multiple lung and myocardial metastasis. A large amount of blood was found in left thoracic cavity. Pathological findings were consistent with PA leiomyosarcoma with ruptured PA aneurysm. It was speculated that the cause of death was rupture of PA aneurysm.

In general, PA sarcoma shows FDG uptake.[2] In this case, PET-CT showed FDG accumulation in multiple lung metastases, but not in pulmonary artery aneurysm. This is because the wall of the aneurysm is too thin for FDG-PET to show FDG uptake. Because of false negatives, therefore, it might be difficult to detect primary pulmonary artery sarcoma with FDG-PET in the presence of an aneurysm.

Two out of 3 patients with main PA aneurysm due to PA sarcoma (2 cases[3,4] have been reported in addition to the present case) experienced rupture of main PA aneurysm. Rupture of PA aneurysm is rare because of low pressure of PA.[5] But PA aneurysm due to PA sarcoma might be fragile and have a higher risk of rupture than main PA aneurysm due to other diseases.

In conclusion, it might be difficult to detect primary lesion of PA sarcoma with FDG-PET in the presence of an aneurysm. More cases are needed to determine the clinical feature of PA sarcoma with main PA aneurysm.

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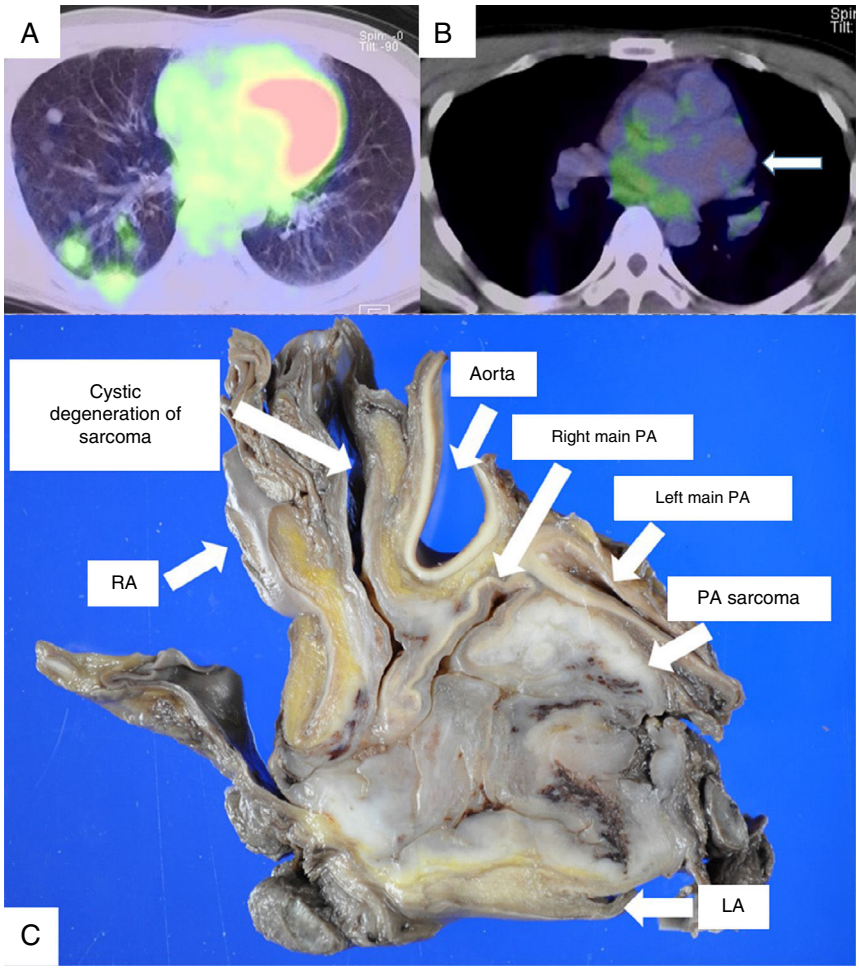

Fig. 1. PET/CT showed FDG uptake in multiple lung nodules (A), but not in PA aneurysm (B). The thin arrow indicates PA aneurysm (B). Macroscopic findings from the specimen obtained by autopsy revealed PA leiomyosarcoma with cystic degeneration (C). PA, pulmonary artery; LA, left atrium; RA, right atrium.

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