

A Case of Sclerosing Hemangioma Evaluated with Diffusion-Weighted Magnetic Resonance Imaging and ^{18}F -Fluorodeoxyglucose Positron Emission Tomography

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A 33-year-old female patient was referred to our hospital for further examination of an abnormal shadow evident on a chest X-ray film. Chest computed tomography (CT) revealed a solid nodule 1.9 cm in diameter in the hilum of the upper lobe of the left lung. Positron emission tomography showed high ^{18}F -fluorodeoxyglucose accumulation in the nodule with a maximal standardized uptake value of 4.5, which favored a malignant lesion. Diffusion-weighted magnetic resonance imaging (DWI), which shows differences in the diffusion of water molecules and can discriminate between malignant and benign lesions, indicated that the nodule had a minimum apparent diffusion coefficient of $1.7 \times 10^{-3} \text{ mm}^2/\text{sec}$, which was higher than the cutoff value of $1.1 \times 10^{-3} \text{ mm}^2/\text{sec}$ for discriminating between malignant and benign diseases; i.e., values equal to or lower than $1.1 \times 10^{-3} \text{ mm}^2/\text{sec}$ favor malignant disease. The results of a CT-guided needle biopsy of the nodule favored sclerosing hemangioma. During surgery, the tumor did not appear to be invasive, and lymph node metastasis and dissemination were not apparent. On the basis of gross appearance, location, preoperative histological diagnosis, and DWI findings, the tumor was enucleated from the pulmonary parenchyma. Seven months after surgery, the patient was alive and had no evidence of recurrent disease. (*Ann Thorac Cardiovasc Surg* 2010; 16: 276–280)

Key words: sclerosing hemangioma, ^{18}F -fluorodeoxyglucose positron emission tomography, diffusion-weighted magnetic resonance imaging

Introduction

Sclerosing hemangioma is a rare lung tumor thought to derive from primitive respiratory epithelium,^{1,2)} as first reported by Liebow and Hubell in 1956.³⁾ Although one case with recurrence⁴⁾ and several with hilar or mediastinal lymph node involvement^{2, 5–9)} have been reported, no

deaths associated with this disorder have been reported.²⁾

In recent years, positron emission tomography (PET) using ^{18}F -fluorodeoxyglucose (FDG) has been successfully used to discriminate malignant from benign pulmonary nodules.^{10–12)} However, FDG-PET has been reported to give false-positive results in some cases of sclerosing hemangioma,^{13,14)} which adversely affects the ability to discriminate between benign and malignant nodules and to develop an appropriate therapeutic strategy.

Recent developments in magnetic resonance (MR) gradient technology have enabled the acquisition of diffusion-weighted MR imaging (DWI), which provides excellent tissue contrast based on differences in the diffusion of water molecules among tissues^{15–17)} and is different from ordinary T₁- and T₂-weighted images. Because the diffusion of water molecules is disturbed by intracellular

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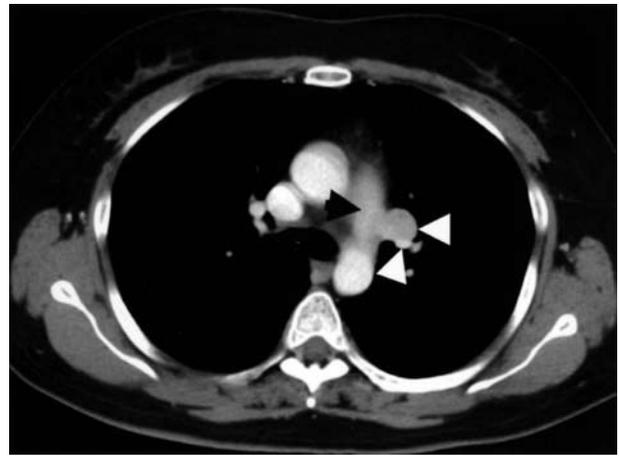
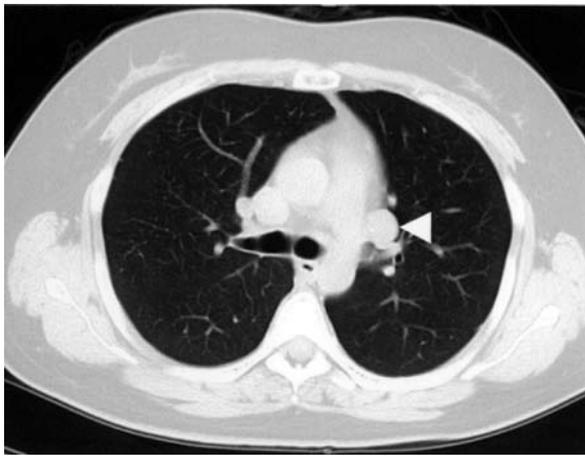


Fig. 1. Enhanced chest computed tomography (a: lung window; b: mediastinal window) revealed a well-circumscribed solid nodule (white arrowhead) 1.9 cm in diameter. The nodule was located in the hilum of the upper lobe of the left lung adjacent to the left pulmonary artery (black arrow) and apicodorsal branch (V^{1+2}) (white arrow) of the left superior pulmonary vein.

a | b

organelles and macromolecules, any structural changes in the proportion of extracellular to intracellular water molecules will alter the signal intensity of DWI and the apparent diffusion coefficient (ADC) of the tissues.^{18–20} Compared with normal tissue, malignant tumors have greater cellularity, larger nuclei with more abundant macromolecular proteins, larger nuclear cytoplasmic (N/C) ratios, and less extracellular space. Therefore the diffusion of water molecules in malignant tumors is restricted relative to that in normal tissue, which results in a lower ADC.^{21–24}

Here we discuss the usefulness of DWI in diagnosing and developing a therapeutic strategy in a case of sclerosing hemangioma with intermediate FDG accumulation.

Case Report

A 33-year-old female patient, who currently was a smoker, was referred to our hospital for further examination of an abnormal shadow on a chest X-ray film found during a general examination in July 2008. Chest computed tomography (CT) revealed a well-circumscribed solid nodule 1.9 cm in diameter, which was located in the hilum of the upper lobe of the left lung, adjacent to the left pulmonary artery and apicodorsal branch (V^{1+2}) of the left superior pulmonary vein (Fig. 1). PET showed high FDG accumulation in the nodule with a maximal standardized uptake value (SUVmax) of 4.5 in the early phase (Fig. 2). The SUVmax is commonly used to discriminate between benign and malignant lung tumors, with a value greater



Fig. 2. ¹⁸F-fluorodeoxyglucose (FDG) positron emission tomography showed high FDG accumulation in the lung nodule (black arrowhead) and a maximal standardized uptake value (SUVmax) of 4.5 in the early phase.

than the cutoff value of 2.5 favoring a malignant lesion.²⁵ In our previous DWI study, we determined the cutoff value of minimum apparent diffusion coefficient (ADCmin) for discrimination between benign and malignant pulmonary nodules to be $1.1 \times 10^{-3} \text{ mm}^2/\text{sec}$. Using this value, we found that DWI showed higher specificity than PET for benign/malignant discrimination.²⁶ In this case, DWI indicated that the nodule had an ADCmin of $1.7 \times 10^{-3} \text{ mm}^2/\text{sec}$, which favored a benign lesion (Fig. 3). The results of a CT-guided percutaneous needle biopsy of the nodule, which was performed by an experienced inter-

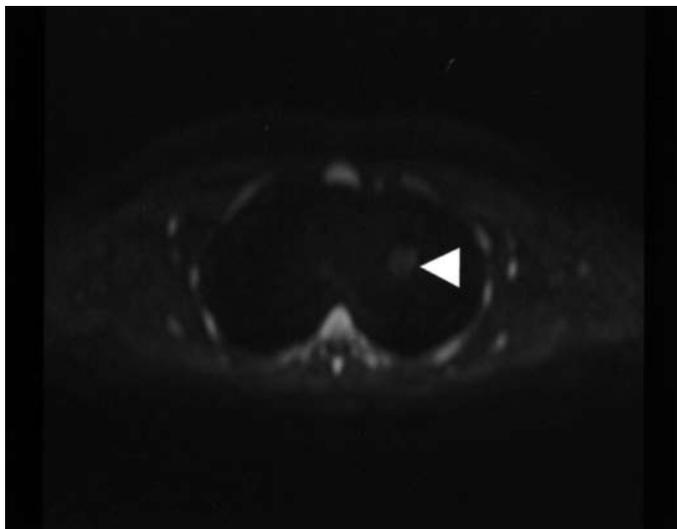


Fig. 3. Diffusion-weighted magnetic resonance imaging showed high intensity in the nodule with a minimum apparent diffusion coefficient (ADC_{min}) of 1.7×10^{-3} mm²/sec. In our previous study, an ADC_{min} value equal to or greater than 1.1×10^{-3} mm²/sec was shown to favor benign disease.

ventional radiologist, favored sclerosing hemangioma. Surgery was performed in September 2008. During thoracoscopic examination of the left thoracic cavity, the nodule was found to be well circumscribed, bulging hemispherically from the surface of the hilum of the upper portion of the lung's left upper lobe. After antero-axillary thoracotomy followed by taping of the apicodorsal (V¹⁺²) and ventral (V³) branches of the left superior pulmonary vein and ventral branch of the left pulmonary artery, the tumor showed no malignant features (i.e., invasiveness or lymph node metastasis or dissemination). On the basis of the tumor's gross features, preoperative histological examination of the biopsy sample, and DWI findings, the tumor was enucleated from the pulmonary parenchyma. A diagnosis of sclerosing hemangioma (Fig. 4) with an MIB-1 index of 1.8% was made after a postsurgical pathological examination of the tumor (Fig. 5). Seven months after surgery, the patient was alive and had no evidence of recurrent disease.

Discussion

FDG-PET is used to discriminate between benign and malignant solitary pulmonary nodules and has a sensitivity and specificity of 96.8% and 77.8%, respectively, as reported by Gould et al.¹²⁾ However, FDG-PET has been reported to give false-negative results for well-differentiated

pulmonary adenocarcinoma^{27,28)} and false-positive results for inflammatory nodules.^{29,30)} Although sclerosing hemangioma is a benign lung tumor, it sometimes shows FDG accumulation on PET,¹⁴⁾ which makes it difficult to distinguish this disease from malignant lung tumors.

DWI discriminates between benign and malignant solitary pulmonary nodules²⁶⁾ in a manner different from that of FDG-PET, which is based on differences in water molecule diffusion between benign and malignant pulmonary nodules. Because the diffusion of water molecules in malignant tumors is restricted by increases in the number of intracellular organelles and macromolecules, any structural changes in the proportion of extracellular to intracellular water molecules will alter the signal intensity of DWI and the ADC of the tissues, resulting in a lower ADC_{min} value for malignant tumors than for benign tumors.¹⁸⁻²⁰⁾

In our previous study, we determined that the ADC_{min} cutoff value for discriminating between benign and malignant nodules was 1.1×10^{-3} mm²/sec.²⁶⁾ A tumor with an ADC_{min} equal to or lower than this value is considered to be malignant. The ADC_{min} value in our patient was 1.7×10^{-3} mm²/sec, which favored benign disease. However, the SUV_{max} value of the tumor was 4.5, which favored malignancy. The results of a CT-guided needle biopsy of the nodule favored sclerosing hemangioma. On the basis of gross features of the tumor, preoperative histological

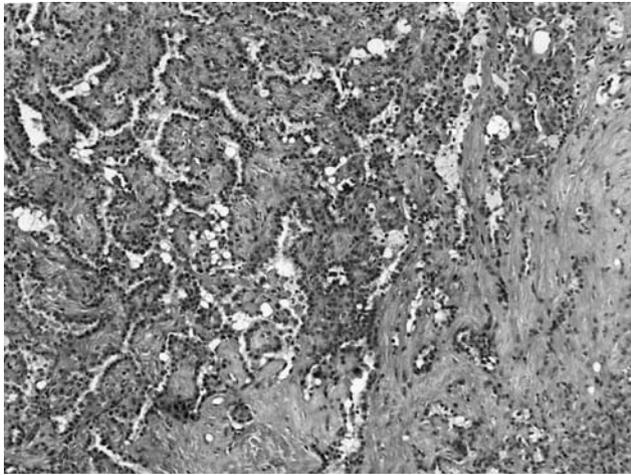


Fig. 4. Findings from a microscopic examination of the lung tumor. The tumor had a sclerotic and papillary pattern.

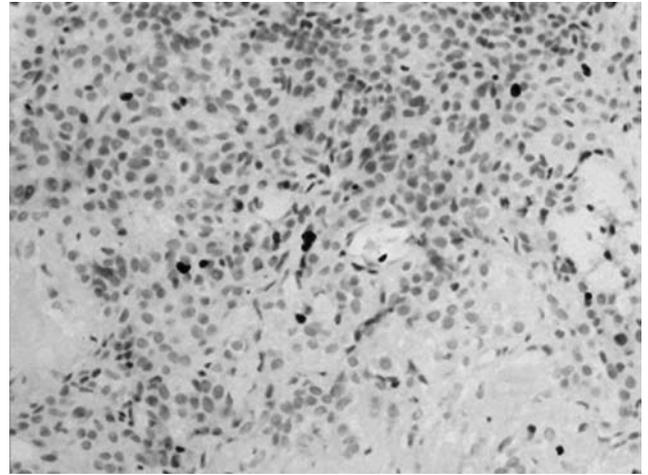


Fig. 5. Findings from an immunohistochemical examination of the lung tumor. The mindbomb homolog-1 positive rate (MIB-1 labeling index) was 1.8%.

examination of the biopsy sample, intraoperative findings of no invasiveness, and DWI findings, the tumor was enucleated from the pulmonary parenchyma.

In our previous study of DWI, a lung schwannoma was positive for FDG accumulation, but it had an ADCmin value higher than the cutoff value²⁶⁾, similar to our findings in the present study for sclerosing hemangioma. We conclude that DWI might compensate for the disadvantages of FDG-PET in discriminating between malignant and benign diseases. Additional studies are needed to clarify the usefulness of DWI in discriminating between malignant and benign lung tumors, especially sclerosing hemangioma.

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