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Surgical Treatment of Idiopathic Multiple Pulmonary Arteriovenous Malformation Identified at the Onset of Cerebral Infarction: A Case Report

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Abstract

Pulmonary arteriovenous malformation (PAVM), which is asymptomatic in most cases, is often identified in patients with central nervous system disorders such as brain abscesses and/or cerebral infarctions. We have reported a patient with idiopathic multiple PAVM identified at the onset of cerebral infarction. A 69-year-old woman visited the Department of Neuropathic Internal Medicine at our hospital with chief complaints of numbness in her left hand and a feeling of weakness. The patient was given a diagnosis of subacute cerebral infarction. Multiple old lacunar infarctions were also observed in the deep white matter of the left frontal lobe. Chest computed tomography showed multiple nodular structures, mainly in the right lower lung field (S8), as well as continuous arteries and veins at the site; thus, the patient was finally diagnosed with multiple PAVM. Right lower thoracoscopic lobectomy was performed, as is typical surgical practice in such cases. The patient had a favorable postoperative course, and had no recurrence of cerebral infarction. Although the patient's lesions were mainly restricted to S8, the fact that there were multiple lesions deemed a lobectomy as the appropriate course of treatment. This case emphasizes that attention should be paid to cases of multiple PAVM since cerebral infarction may arise from the disease.

Keywords

Pulmonary Arteriovenous Malformation (PAVM), Brain Abscesses, Cerebral Infarctions

1. Introduction

Pulmonary arteriovenous malformation (PAVM), which is asymptomatic in most cases, is often identified in patients with central nervous system disorders such as brain abscesses and/or cerebral infarctions, in addition to patients with blood-stained sputum and hypoxemia-induced dyspnea or polycythemia. In Europe and the United States, hereditary hemorrhagic telangiectasia (HHT) is concurrently found in 60% - 80% of patients [1] [2]. The mortality rate of this disease is high, at around 10%, and aggressive treatment such as interventional radiology (IVR) and surgery is recommended immediately after diagnosis [3]. We describe our experience with a surgical case of idiopathic multiple PAVM which was identified at the onset of cerebral infarction.

2. Case

A 69-year-old woman visited the Department of Neuropathic Internal Medicine at our hospital with chief complaints of numbness in her left hand and a feeling of weakness. Although the patient had previously experienced a few atonic seizures, she had recovered in 1 to 2 hours and did not visit the hospital. The patient had no history of recurrent epistaxis, and was 153 cm in height and 46 kg in weight. There was no heart murmur, and respiratory sound was clear. There were no abnormal findings in her chest and abdomen and no capillary dilatation in her skin or mucosa. The patient did not exhibit quadriplegia. Magnetic resonance imaging taken at the first visit showed high signal intensities in the right centrum semiovale and the precentral gyrus on the diffusion-weighted images, and high signal intensities were also seen on fluid attenuated inversion recovery images. The patient was given a diagnosis of subacute cerebral infarction. Multiple old lacunar infarctions were also observed in the deep white matter of the left frontal lobe (**Figure 1**). Chest computed tomography showed multiple nodular structures, mainly in the right lower lung field (S8), as well as continuous arteries and veins at the site; thus, the patient was finally diagnosed with multiple PAVM (**Figure 2** and **Figure 3**). Although IVR was first considered, multiple, relatively widespread lesions were observed in the peripheral lung field; therefore, right lower thoracoscopic lobectomy was performed, as is typical surgical practice in such cases. The patient had a favorable postoperative course, and had no recurrence of cerebral infarction. Pathological examination revealed no neoplastic change, and the diagnosis of multiple PAVM was confirmed (**Figure 4**). There were small and large arteriovenous lesions along the bronchus leading to the lower lobe, primarily in S8. Three years after surgery, no signs of recurrence have been observed.

3. Discussion

PAVM is a congenital or acquired abnormal intrapulmonary arteriovenous shunting. In Europe and the United States, 80% of patients with PAVM are reportedly congenital in nature, and 47% - 80% have HHT, which is an autosomal dominant inheritance [1] [4]. Although some cases of PAVM are caused by

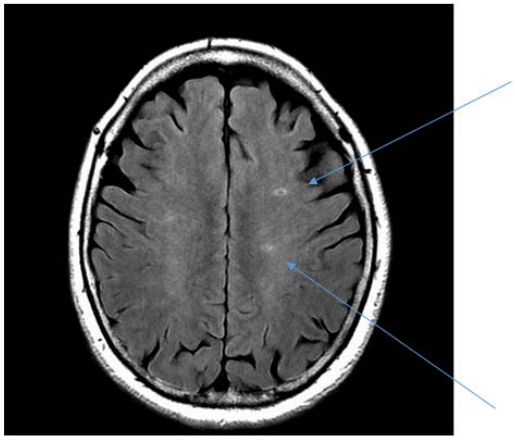

Figure 1. Magnetic resonance imaging (DW2 FLAIR). Multiple old lacunar infarctions were also observed in the deep white matter of the left frontal lobe. Arrows are acute infarct lesions.

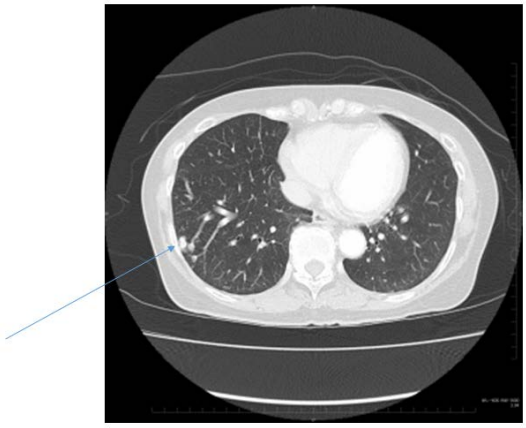

Figure 2. Chest computed tomography. Chest computed tomography showed multiple nodular structures, mainly in the right lower lung field (S8), as well as continuous arteries and veins at the site; thus, the patient was finally diagnosed with multiple PAVM.

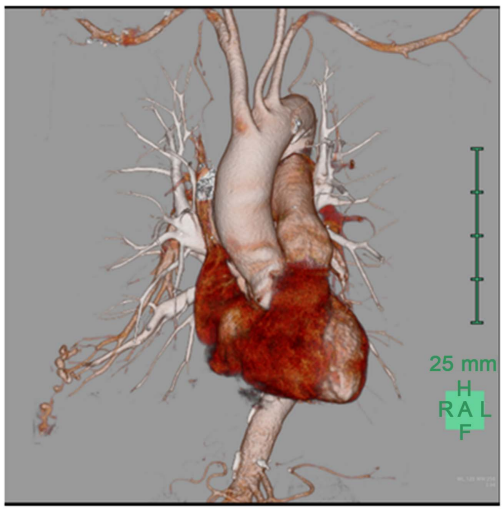

Figure 3. CT angiography. Nodule-like structures occur frequently in the vicinity of the bronchial bifurcation and continue with the peripheral pulmonary artery and vein.

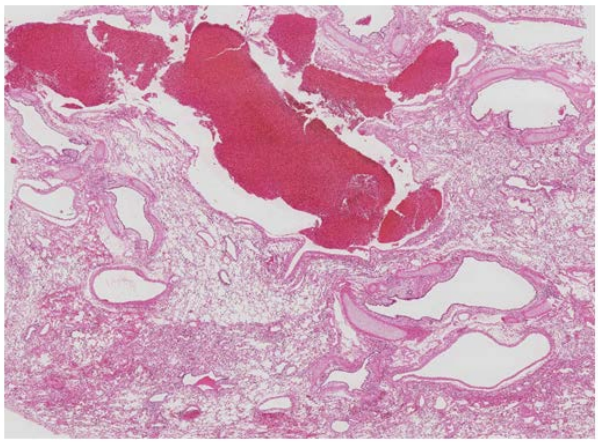

Figure 4. Pathological findings. Pathological examination revealed no neoplastic change, and the diagnosis of multiple PAVM was confirmed. Abnormal vessels spreading to the vicinity of the bifurcation of the lower bronchial lobe are recognized.

trauma, liver cirrhosis, paragonimiasis, and malignant tumors [5], the case presented here was considered idiopathic because the patient presented with no symptoms suggestive of HHT or other causes such as recurrent epistaxis, capillary dilatation in the skin and mucosa, or arteriovenous malformations of the lung, liver, brain, gastrointestinal tract, or spinal cord.

The symptoms of PAVM are often nonspecific and respiratory in nature. Such symptoms include respiratory discomfort, cough, chest pain, malaise, and blood-stained sputum. In addition, the central nervous system is affected in approximately 30% of patients in the form of stroke, transient ischemic attacks, and brain abscesses [1] [2]. The mechanisms underlying brain abscesses and cerebral infarctions include: 1) decreased resistance to infection in brain tissues due to hypoxemia; 2) development of ischemia or infarction in the brain due to polycythemia-induced embolizationa; and 3) septic emboli, often reaching the brain through the fistula [6]. The incidence of cerebral infarction is reportedly 32% in single PAVM and 60% in multiple PAVM [7]. This case was also found in the symptoms of repeated cerebral infarction.

Treatment of PAVM is indicated for a size cut-off of ≥ 3 mm for the feeding artery when the risk of cerebral infarction is high [8]. Although IVR is commonly the first-line therapy [9], and about 16% of patients experience recurrence [10]. Moreover, the following complications have been reported: thrombosis, cerebral infarction by coil migration, brain abscesses (the left cardiac system to the brain), coil protrusion into the aorta, rupture of a fragile venous sac, outflow of arteriovenous thrombi, and pulmonary infarction caused by obstruction of the pulmonary artery bifurcating to the normal lung. Therefore, surgical resection is recommended for patients with multiple pulmonary arteriovenous malformations, such as our patient presented here. Thoracoscopic surgery has been performed in many institutions over recent years [11]. However, in order to recommend such surgery, the lesion needs to be benign and the extent of resection should be as small as possible; therefore, partial resection is commonly per-

formed. In the present case, since lesions were spread around the bronchus in S8 to the lower lobe bronchus, a lobectomy was considered appropriate.

4. Conclusion

We have reported a patient with idiopathic multiple PAVM identified at the onset of cerebral infarction. Although the patient's lesions were mainly restricted to S8, the fact that there were multiple lesions deemed a lobectomy as the appropriate course of treatment. This case emphasizes that attention should be paid to cases of multiple PAVM since cerebral infarction may arise from the disease.

Consent

Written informed consent was obtained from the patients for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in Chief of this journal

Authors Contribution

MK have operated this case and analyzed all data. SY, NT, KI, SH and YO did the assistant of the operation. All authors read and approved the final manuscript.

Competing Interests

The authors declare that they have no competing interests.

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