

# Pulmonary Arteriovenous Fistula with Cerebral Infarction Successfully Treated By Video-Assisted Thoracic Surgery

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We experienced a case in which a pulmonary arteriovenous fistula was found due to the occurrence of cerebral infarction. The patient was a 65-year-old female who had seen a local doctor for lightheadedness occurring upon rising in the morning. She underwent a cerebral MRI and was diagnosed to have a cerebral infarction. Upon closer examination, a chest radiograph revealed an abnormal shadow and a three-dimensional computed tomography (3D-CT) chest angiography detected a pulmonary arteriovenous fistula 32×30 mm in size with the feeder blood vessel A<sup>10</sup> and drainer blood vessel V<sup>10</sup> in the left inferior lobe S<sup>10</sup>, which was considered to be the cause of the cerebral infarction. Video-assisted thoracic surgery (VATS) was conducted for a segmental resection of the basal segment of the left lung. The patient's postoperative progress was good and there was no reoccurrence of the cerebral infarction. The rate of occurrence of cerebral infarction along with pulmonary arteriovenous fistula is considered to be 11.4%; however, cerebral infarction may be an early indicator of pulmonary arteriovenous fistula, and therefore, due attention must be paid. (*Ann Thorac Cardiovasc Surg* 2008; 14: 35–37)

**Key words:** arteriovenous fistula, cerebral infarction, video-assisted thoracic surgery

## Introduction

Most pulmonary arteriovenous fistulae have no symptoms and are detected as an abnormal shadow in the chest, with a conclusive diagnosis being made by means of pulmonary arteriography or three-dimensional computed tomography (3D-CT) angiography. While it is known to have various complications, we experienced a case of pulmonary arteriovenous fistula with the occurrence of cerebral infarction, and performed video-assisted thoracic surgery (VATS).

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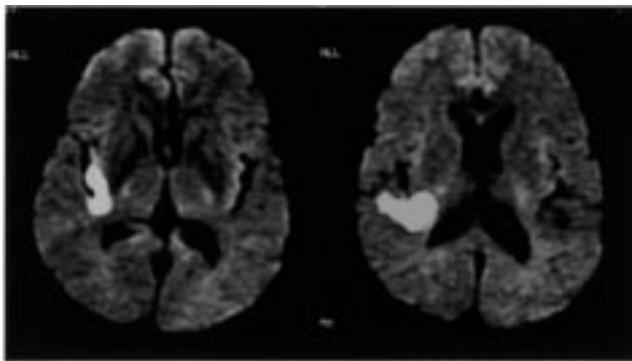
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## Case Report

A 65-year-old female visited a local clinic for lightheadedness occurring upon standing. An MRI diffusion weighted image of the head found a lesion across the cortex and cinerea, and the patient was diagnosed to have a cerebral infarction resulting from an obstructing lesion in the peripheral middle cerebral artery (Fig. 1). Upon closer examination, a chest radiograph revealed an abnormal shadow in the lower left lung region where the cardiac shadow overlaps (Fig. 2), and a 3D-CT angiography of the chest detected a pulmonary arteriovenous fistula 32×30 mm in size with the feeder blood vessel A<sup>10</sup> and drainer blood vessel V<sup>10</sup> in the left inferior lobe S<sup>10</sup> (Fig. 3). The pulmonary arteriovenous fistula was considered to be the cause of the cerebral infarction. Neither cyanosis nor finger clubbing was observed. Furthermore, no abnormalities were found in blood and biochemical tests, and neither polycythemia nor hypoxemia were



**Fig. 1.** Brain MRI.

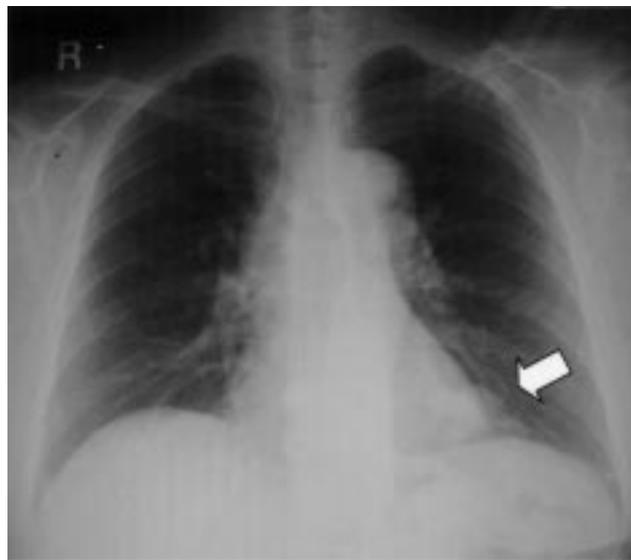
A diffusion weighted image found a lesion across the cortex and cinerea diagnosed to be a cerebral infarction resulting from an obstructing lesion in the peripheral middle cerebral artery.

found.

Surgery was performed based on diagnosis of the single isolated left pulmonary venovenous fistula. VATS was conducted in the lateral decubitus position with the right side down for segmental resection of the basal segment of the left lung with an access thoracotomy of approximately 8 cm in the fifth intercostal space of the chest (Fig. 4). In an isolated preparation, the size of the lesion was found to be 36×32 mm with A<sup>10</sup> and V<sup>10</sup> passing through the incision plane where the saclike enlargement was observed. The diameter of drainer blood vessel was 5 mm. A histopathological examination found closely aggregated arteries and veins with enlarged lumens showing abnormal structure, and sporadically occurring capillary proliferation along with fibrotic thickening in the alveolar interstitium, and thus, it was diagnosed to be a pulmonary arteriovenous fistula. The patient's postoperative progress was good, and there was no reoccurrence of cerebral infarction.

## Comment

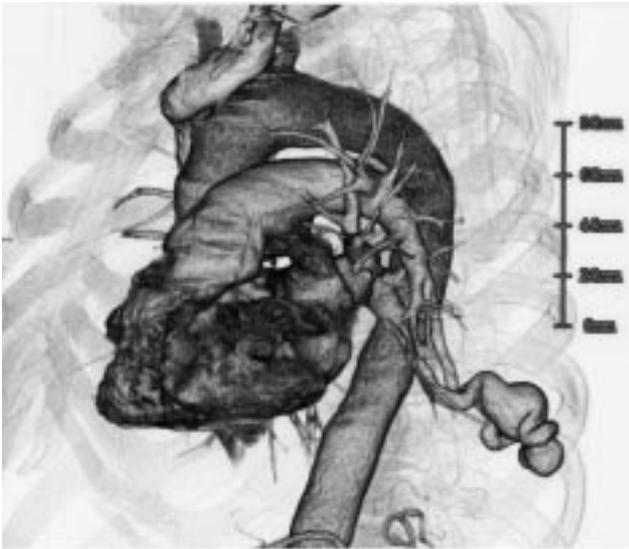
Pulmonary arteriovenous fistula is a relatively rare disease, considered to occur at a frequency of 0.02%.<sup>1)</sup> Most cases are congenital and considered to be abnormal developments of the capillaries, and cases associated with Rendu-Osler-Weber have also been reported.<sup>1,2)</sup> Twenty-eight percent of cases are considered to have no symptoms.<sup>1)</sup> Dines et al. reported that when a single isolated pulmonary arteriovenous fistula is 2 cm or smaller, no symptoms appear.<sup>3)</sup> Generally, when the right-left shunt



**Fig. 2.** Chest Xp.

A chest radiograph revealed an abnormal shadow found in the lower left lung region where the cardiac shadow overlaps.

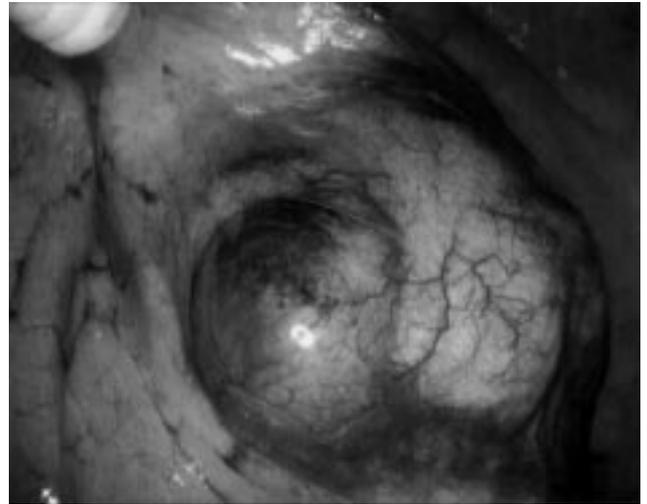
is 20–30% or greater, symptoms such as breathing difficulty, cyanosis, hypoxemia, finger clubbing and polycythemia occur, and the rate of occurrence of severe complications such as rupture of the fistula, hemoptysis, cerebral infarction and cerebral abscess is considered to be approximately 30%.<sup>1)</sup> As for the cerebral complications seen in this case, the rate of occurrence is considered to be 6.8% for cerebral abscess and 11.4% for cerebral infarction, with a fatality rate as high as 23%.<sup>1)</sup> A cerebral infarction is thought to occur by the following three mechanisms: (i) embolization caused by thrombus formation in a fistula, (ii) embolization caused by deep-vein thrombosis in the inferior limb, and (iii) thrombus or embolization caused by polycythemia. In this case, neither deep-vein thrombosis nor polycythemia was observed; therefore, (i) the mechanism was most likely. Paradoxical embolization tends to occur particularly in a fistula of a feeder artery with a diameter over 3 mm in a right-left shunt,<sup>4,5)</sup> so in that regard, this case required care because the diameter was as large as 5 mm. In terms of treatment, surgical removal including thoracoscopic surgery<sup>6)</sup> and transcatheter embolization<sup>4,5)</sup> can be performed. Although no symptoms appear, there is a possibility of continuous growth and future complications, so treatment is recommended. In fact, the death rate of patients with untreated pulmonary arteriovenous fistula for an average of 8 years was reported to be 22.2% (6 of 27 cases).<sup>3)</sup>



**Fig. 3.** Chest 3D-CT angiography.

A pulmonary arteriovenous fistula 32×30 mm in size with the feeder blood vessel A<sup>10</sup> and drainer blood vessel V<sup>10</sup> was found in the left inferior lobe S<sup>10</sup>.

Treatment should be chosen depending on such factors as size, region and quantity of the fistula. Although transcatheter embolization is less invasive, recurrences caused by migration of the embolism, rupture of the fistula, or recanalization have been reported.<sup>4)</sup> Owing to recent developments in diagnostic imaging, pulmonary arteriovenous fistulae with small diameters have been detected more frequently, and opportunities for surgical removal have thus increased due to the effectiveness of thoracoscopic surgery. As seen in this case, a cerebral infarction may appear in patients as a preliminary indication of pulmonary arteriovenous fistulae, so it is considered to be a disease that requires due attention.



**Fig. 4.** Intraoperative thoracoscopic findings.

In the left inferior lobe S<sup>10</sup>, a saclike enlargement was observed which was then determined to be a pulmonary arteriovenous fistula.

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