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Intracerebral Hemorrhage after Lung Lobectomy in Patients with Congenital Pulmonary Artero-Venous Malformation (AVM)

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ABSTRACT

A 13-year-old boy was diagnosed with isolated congenital pulmonary AV fistula at the left lower lobe of the lung. He was operated for left lower lobe lobectomy. 18 hours after the operation, the patient developed intracerebral and intraventricle hemorrhage with hydrocephalus and brain edema. As a result he was operated on again for a ventriculoperitoneal shunt. 5 months after the operation. Brain MRI was done and indicated a suspected AVM at pineal region. 6 months after the operation, he reported normal health condition and could regularly perform daily activities including exercises.

Keywords: AVM; intracerebral hemorrhage; lobectomy

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A thirteen years old Thai male reported an unhealthy heart condition since the age of seven years old while he had no history of any diseases and was healthy prior to seven years old of age. The boy's health was reported to be worse as time went on. He now could not exercise and could not comfortably walk over 50 meters distance. His medical examinations detected central cyanosis and clubbing of fingers, no telangiectasia, soft gr 2/6 systolic murmur at left lower chest, and normal heart sound but no heart murmur. In addition, his heart was detected 73.4% Hct and 79-80% O_2Sat, and the chest X-ray (CXR, Fig 1) displayed a soft tissue mass with 5 cm diameter. His cardiac catheterization pulmonary angiogram also showed a large pulmonary AV fistula at the left lower lobe of the lung, meanwhile heart condition was found normal after echocardiography performance.

The lower left lobe lobectomy detected the large AVM in the boy's left lower lobe lung with the feeding vessel of about 1.5 cm in diameter branching from the pulmonary artery (Fig 2).

After operation, the boy was found reported as 95% O_2Sat, could move his body and ET tube was removed within 6 hours post operation. However, after eighteen hours post operation, the patient became unconscious and numb at both arms and legs. Consequently, he was re-intubated and CT brain scanned, and Fig 3 showed the results of intracerebral and intraventricle hemorrhage with hydrocephalus and brain edema.



FIG 1. Patient's CXR before operation.

Next, the patient was operated on for ventriculoperitoneal shunt and improvement of symptoms was reported. Brain MRI was performed at five months after operation, and indicated the potential reason for intracerebral hemorrhage from a large amount of old bleeding at the

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Fig 2. AVM tissue at the lower left lobe lung.

pineal region underlying the AVM.

At six months after operation, he reported normal health conditions and could regularly perform daily activities including exercises.

DISCUSSION

To date, studies have rarely found the case of intracerebral hemorrhage after the lung lobectomy AVM. Nonetheless, within the particular group of patients with the lung AVM cases, intracranial AVM was often observed in the hereditary hemorrhagic telangiectasia group of patients.

Hereditary Hemorrhagic Telangiectasia (HHT) or Osler-Weber-Rendu (OWR) is defined as multisystem vascular dysplasia, leading to multiple AVM at various organs. The HHT type 1 gene mutation is caused by the mutation of endoglin gene (chromosomal locus 9q34), while the HHT type 2 mutation is caused from the mutation of ALK1 gene (chromosome locus 12q1). In general, HHT is diagnosed through following clinical diagnosis rules. Definite HHT is considered when three or more of the criteria below are present, a possible or suspected case is when two of the criteria below are present, and an unlikely case is when fewer than two of the criteria below are present.

Diagnosis criteria for HHT

1. Nose bleeds (epitasis): spontaneous and recurrent.
2. Telangiectases: multiple, at characteristic sites, including face, lips, oral cavity and fingers.
3. Visceral AVM (pulmonary, cerebral, hepatic, spinal) or gastrointestinal telangiectases (with or without bleeding)

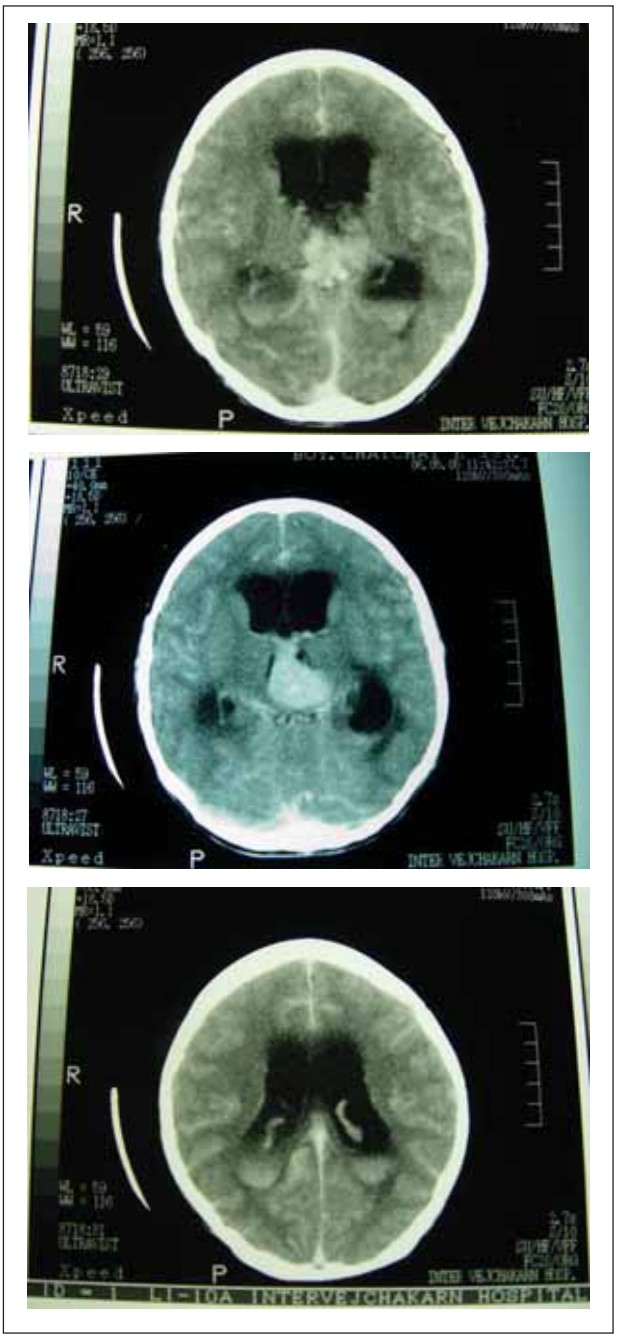


Fig 3. CT image of the patient's one day after lobectomy surgery.

4. Family history: a first degree relative with HHT according to these criteria.

In conclusion, the 13 years old boy visited the hospital with a large AVM in his lung. Since he and his family had no history of this HHT disease, his AVM in other organs were not examined before the lobectomy surgery. Nevertheless, intracerebral hemorrhage was detected after the first operation. This indicates the importance of brain AVM examination in all cases, especially in patients who will undergo lung AVM surgery.

For the patients with a small AVM in the lungs, transcathether embolization treatment generally provides a safe and suitable method of treatment, whereas transcathether embolization does not work for patients with a large AVM. Thus, the operation remains the main method of treatment, given that the doctors and/or the clinicians must be aware of any possible side effects via intracerebral hemorrhage as reported in this study.

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