Coil Embolization for Pulmonary Arteriovenous Malformation as an Organ-sparing Therapy: Outcome of Long-term Follow-up

Koji Ando, MD,1 Atshushi Mochizuki, MD,1 Noriaki Kurimoto, MD,1 Kumio Yokote, MD,1 Yasuo Nakajima, MD,2 Hiroaki Osada, MD,3 and Haruhiko Nakamura, MD1

Purpose: Pulmonary artery coil embolization (PACE) is increasingly utilized to treat pulmonary arteriovenous malformations (PAVMs), but the long-term outcome of this treatment modality remains unclear. By evaluating the long-term outcome of patients at St. Marianna University Hospital treated with PACE, we wanted to see if PACE could effectively replace the surgical resection of PAVMs.

Patients and Methods: We retrospectively evaluated 9 consecutive patients (4 males, 5 females; age range, 16–67 years; mean ± SD, 43.6 ± 18.7 years) who underwent PACE for PAVMs. Selective pulmonary artery angiography using Seldinger’s method was initially performed to identify the feeding arteries. This was followed by embolization using interlocking detachable coils and microcoils.

Results: The procedure resulted in no severe complications. All treated patients were free from PAVM symptoms. A recurrence did not occur after PACE in 8 of 9 (88%) patients for 31 to 173 months (86 ± 51). Recanalization of the embolized malformation occurred after 3 months in one patient. This patient underwent an additional successful PACE without any further recurrences for 73 months.

Conclusions: PACE is an organ-sparing therapy with satisfactory long-term results. It can safely replace the surgical resection of PAVMs.

Key words: arteriovenous malformation, coil embolization, intervention, malformation, pulmonary vessel

Introduction

A pulmonary arteriovenous malformation (PAVM) is a relatively rare congenital disease caused by abnormal communications between the pulmonary arteries and pulmonary veins.1) About 70% of patients with PAVMs in western countries have hereditary hemorrhagic telangiectasia, which is transmitted in an autosomal dominant inheritance.2) The rate is lower in Asians than in people of European descent. Patients with PAVMs usually have multiple lesions as a systemic disease. The malformations are made of thin-walled vascular channels and connective tissue stroma. A PAVM lesion may be a large single sac, a plexiform mass of dilated vessels, or a tortuous direct communication between arteries and veins. Depending on the size and number of lesions, symptoms vary from totally absent to severe dyspnea with cyanosis due to right-to-left blood shunt.3) Dissemination of thrombi to the brain causes brain abscesses, especially for the hereditary type of PAVM.4) Rupture of a large
PAVM can induce a life-threatening massive pulmonary hemorrhage. Therefore, curative treatment is necessary, even for subclinical cases.

Surgical resection or exteriorization of a vascular fistula has long been the therapy for PAVMs. Minor lung resection, such as excisional resection or wedge resection, may be enough for a cure when a PAVM is solitary, small, and located in the peripheral regions of the lungs. However, a larger resection (including lobectomy or segmentectomy) is often necessary, depending on the size, number, and location of lesions.

Embolotherapy—including balloon embolization and pulmonary artery coil embolization (PACE)—has been increasingly utilized recently to treat PAVMs. As an organ-sparing therapy, it is initially successful, but the long-term outcome of this treatment modality needs to be determined. By evaluating retrospectively the long-term results of PACE performed at St. Marianna Hospital, we could determine whether PACE can replace surgery as a treatment for PAVMs.

**Patients and Methods**

**Patients**

Between June 1993 and December 2004, nine consecutive patients with PAVMs were treated with PACE at St. Marianna University Hospital. They were 4 males and 5 females and ranged in age from 16 to 67 years (mean \( \pm \) SD, 44 \( \pm \) 19). The presenting signs or symptoms detected during physical check-up were dyspnea (1 patient), continuing dry cough (1 patient), and nodular shadows on chest radiographs (7 patients). The patients’ Karnofsky performance status (PS) ranged from 0 to 1. All patients received PACE to improve their presenting respiratory symptoms or to prevent future undesired events such as enlargement of the malformation, massive pulmonary hemorrhage, brain abscesses, or dyspnea. Prior to PACE, the patients underwent contrast-enhanced chest computed tomography (CT) and pulmonary artery angiography to identify the size, number, location and feeding arteries of the PAVM. The mean follow-up period of the patients was 89 months.

**Pulmonary artery coil embolization**

After applying local anesthesia around the right femoral vein, and by using Seldinger’s method, we inserted a catheter toward the right ventricle for placement of the coil. Selective pulmonary artery angiography was used to measure the size of the feeding arteries. Then, one or two interlocking detachable coils (IDC) (Boston Scientific, Boston, MA) were inserted into the malformation. The coils had a slightly larger diameter than that of the feeding artery. Then, fluorography was used for the additional delivery of multiple Tornade® microcoils (Cook Medical, Bloomington, IN) that completely embolized the extended nidus (Fig. 1A and 1B). The size of the IDCs ranged from 2 to 30 mm, and the size of the Tornade® microcoils ranged 3 to 10 mm. Angiography confirmed the occlusion of the feeding arteries at the end of the procedure. During the procedure, ECG monitoring was continuous, and the patients received intravenous fluids and antibiotics to prevent infection.

**Follow-up of the patients**

The routine follow-up evaluations of the patients took place at the outpatient department after discharge. Every
Ando K et al.

Ann Thorac Cardiovasc Surg Vol. 17, No. 2 (2011)

three months for one year, we evaluated the patients’ chest radiographs and contrast-enhanced chest CTs to detect enlargement or recanalization of the embolized PAVMs. After one year, patients had a routine follow-up evaluation every 6 months or annually.

Statistics

Differences between the PaO2 values before and after PACE were analyzed using the Student’s t-test. A p value <0.05 was considered significant.

Results

Profiles of the patients and the long-term follow-up results are shown in Table 1. Two patients had multiple lesions and seven patients had a single lesion. Only one patient (No. 6) had Osler-Weber-Rendu disease. There were no severe complications resulting from the procedure (Fig. 2A–2C). Dislocation of the coils occurred in two patients (No. 1 and No. 2) during the procedure. The microcoil in the right ventricle unexpectedly dislodged in the first patient and migration of the microcoil to the gastroduodenal artery occurred in the second patient. The dislodged coil in the right ventricle was immediately removed through the catheter, but the migrated coil in the abdominal artery was allowed to remain at the site because the patient had no symptoms and no further migration occurred.

The arterial oxygen partial pressure (PaO2) improved dramatically after PACE therapy in 6 patients. The PaO2 was not measured in 3 patients. The average PaO2 in patients before treatment was 70.3 ± 19.0 Torr. This increased to 86.6 ± 20.8 Torr after PACE therapy (p <0.03, Fig. 3).

Most patients were discharged from the hospital uneventfully one to two days after treatment. Two patients (No. 1 and No. 2) had multiple PAVMs and required a longer hospital stay to undergo 2 to 3 staged PACE treatments. Recanalization of the PAVM occurred in one patient (No. 3) after 3 months. This patient received additional microcoils as a final embolization. Two patients (No. 1 and No. 3) died from other diseases at 72 months and 137 months after discharge. All patients were free from PAVM symptoms and recurrences did not occur from 31 to 173 months after PACE therapy (the average was 85.9 ± 50.7 months).

Discussion

PAVM usually generates a right-to-left blood shunt in which pulmonary oxygenation of the blood does not occur, resulting in hypoxemia and cyanosis. Surgical resection of the arteriovenous fistula can be the most radical therapy. However, a complete resection is sometimes

<table>
<thead>
<tr>
<th>Patient number</th>
<th>Age</th>
<th>Sex</th>
<th>Location</th>
<th>Maximum diameter (mm)</th>
<th>Number of coils used</th>
<th>Number of microcoils</th>
<th>PaO2 (mmHg) before embolization</th>
<th>PaO2 (mmHg) after embolization</th>
<th>Follow-up period (months)</th>
<th>Recanalization of PAVM or associated symptoms</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>67</td>
<td>F</td>
<td>BLLs</td>
<td>25</td>
<td>6</td>
<td>6</td>
<td>46.9</td>
<td>60.1</td>
<td>72</td>
<td>None</td>
</tr>
<tr>
<td>2</td>
<td>56</td>
<td>F</td>
<td>BLLs+LS5</td>
<td>12</td>
<td>3</td>
<td>3</td>
<td>63.7</td>
<td>66.8</td>
<td>173</td>
<td>None</td>
</tr>
<tr>
<td>3</td>
<td>58</td>
<td>F</td>
<td>RS3</td>
<td>30</td>
<td>1</td>
<td>1</td>
<td>ND</td>
<td>ND</td>
<td>172</td>
<td>Recanalization occurred 3 months after the initial PACE necessitating an additional PACE</td>
</tr>
<tr>
<td>4</td>
<td>60</td>
<td>M</td>
<td>RS1</td>
<td>35</td>
<td>1</td>
<td>1</td>
<td>63.6</td>
<td>80.5</td>
<td>54</td>
<td>None</td>
</tr>
<tr>
<td>5</td>
<td>34</td>
<td>M</td>
<td>LS6</td>
<td>15</td>
<td>1</td>
<td>1</td>
<td>78.7</td>
<td>95.5</td>
<td>54</td>
<td>None</td>
</tr>
<tr>
<td>6</td>
<td>44</td>
<td>F</td>
<td>RS6</td>
<td>28</td>
<td>1</td>
<td>1</td>
<td>65.7</td>
<td>106.7</td>
<td>72</td>
<td>None</td>
</tr>
<tr>
<td>7</td>
<td>16</td>
<td>M</td>
<td>RS6</td>
<td>30</td>
<td>1</td>
<td>1</td>
<td>ND</td>
<td>ND</td>
<td>96</td>
<td>None</td>
</tr>
<tr>
<td>8</td>
<td>41</td>
<td>F</td>
<td>RS2</td>
<td>10</td>
<td>1</td>
<td>1</td>
<td>ND</td>
<td>ND</td>
<td>31</td>
<td>None</td>
</tr>
<tr>
<td>9</td>
<td>16</td>
<td>M</td>
<td>LS3</td>
<td>17</td>
<td>1</td>
<td>1</td>
<td>103.1</td>
<td>110.0</td>
<td>73</td>
<td>None</td>
</tr>
</tbody>
</table>

BLLs, bilateral lower lobes; F, female; FA, feeding artery; L, light; M, male; PACE, pulmonary artery coil embolization; PAVM, pulmonary arteriovenous malformation; R, right; S, segment; ND, not determined
Coil Embolization for PAVMs

A: The pulmonary angiography shows an arteriovenous malformation in the right pulmonary segment (S6) in patient No. 6 who had dyspnea due to right-to-left shunt. 
B: In this patient, one interlocking detachable coil and additional multiple microcoils applied to the feeding artery completely abolished the blood flow. Dyspnea due to shunt effects disappeared immediately after the embolization.  
C: A chest radiograph taken 48 months after the arterial embolization shows a calcified lesion at the embolized area.

Fig. 3  A comparison of the average arterial oxygen partial pressure of the patients before (70.3 ± 19.0 Torr) and after (86.6 ± 20.8 Torr) coil embolization (p <0.03). The significant improvement resulted from abolishing the right-to-left blood shunt created by the malformations.
difficult for multiple lesions. In addition, when a major lung resection (including lobectomy) is required, a postsurgical loss of lung function may become a problem. 

Since the early 1980s, interventional therapy by embolization is gradually replacing surgery. 

The angioarchitecture of the malformation is important for embolization because a cure requires the occlusion of all feeding arteries. The treatment indication for asymptomatic PAVM is still unclear. However, several authors have recommended embolotherapy for lesions with a feeding artery diameter of greater than 3 mm to prevent further complications. The diameter of the feeding artery in patients with PAVM was larger than 3 mm and ranged between 4 to 10 mm in size. 

The recurrence of a fistula after PACE is a major concern about the efficacy of this treatment modality. In a long-term follow-up study, Lee et al. reported that 52 large PAVMs in 38 of 45 patients (84%) were cured by the first embolotherapy. Persistent PAVMs in the remaining patients were successfully treated by a second or a third procedure. In this study, one patient (12%) had a recurrence, but an additional one-time embolization cured it. Thus, embolotherapy seems to obtain permanent occlusion of a PAVM in the majority of patients, and patency due to recanalization or accessory artery growth can be easily detected and treated. 

Mager et al. reported that periprocedural complications of embolotherapy occurred in 8% of sessions in 112 patients with PAVMs. Adverse events after embolization were migration of an embolic device, transient ischemic attack (TIA), angina pectoris, and early cerebral infarction. In these patients, dislocation of microcoils occurred during the procedure, but no serious complications were observed during each of the 3-month follow-up periods. 

In summary, since PAVMs did not recur after PACE therapy in 8 of 9 (88%) patients during the long-term follow-up period, we believe this treatment modality can replace surgical resection as an organ-sparing treatment in most patients. PACE for PAVMs is efficacious and tolerable in the majority of patients. However, as we demonstrate, routine follow-up is required since the recanalization of PAVMs or enlargement of untreated PAVMs occurring after treatment cannot be neglected for many years.

References