Combined endovascular and video-assisted thoracoscopic procedure for treatment of a ruptured pulmonary arteriovenous fistula: Case report and review of the literature

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A 35-year-old woman with a medical history of hereditary and recurrent epistaxis and lip telangiectasia was admitted with sudden left thoracic pain and dyspnea. There was no recent history of fever or thoracic trauma. Biologic data were as follows: hemoglobin level, 8.8 g/L; hematocrit level, 28%; oxygen saturation, 97%; and fraction of inspired oxygen, 10 L/min. Chest radiography and computed tomographic (CT) scanning revealed a left hemothorax with a suspicion of a vascular malformation in the left lower lobe (Figure 1). There were no aortic or pericardial abnormalities.

An intercostal drain was placed in the left pleural cavity, and 2000 mL of blood was drained, leading to a significant clinical improvement. Pulmonary angiography confirmed the presence of 2 arteriovenous fistulas in the upper part of the left lower lung and in the lingula (Figure 2). Rendu-Osler-Weber syndrome (ROW) was diagnosed on the basis of the patient's history and clinical and radiologic data.

Embolization of the lower lobe fistula was performed during general anesthesia with 3 Guglielmi Detachable Coils (Boston Scientific, Target Therapeutics, Fremont, Calif). One 3-dimensional shaped coil of 8 × 20 mm and 2 fibered coils of 6 × 20 mm and 4 × 20 mm, respectively, were placed with a 0.018-inch microcatheter (Boston Scientific, Target Therapeutics) in the feeding artery. Occlusion was completed with the injection of 0.3 mL of embucilate (Histoacryl; Braun Aesculap, Tuttlingen, Germany) emulsified with iodized oil (Lipiodol; Guerbet, Aulnay-sous-Bois, France). Catheterization of the lingula fistula was technically more difficult, and occlusion was achieved with a 4 × 20-mm fibered

spiral placed with a 0.018-inch microcatheter (Boston Scientific; Figure 3).

Immediately after the endovascular procedure, video-assisted thoracoscopic surgery was performed to remove any residual clot and exclude further bleeding. The patient was placed in the right decubitus position. A single-lumen endotracheal tube was inserted, and ventilation was performed with reduced tidal volume throughout the procedure. A 10-mm trocar was introduced through a skin incision into the fifth intercostal space in the midaxillary line for insertion of a 0° endoscope (Karl Storz, Tuttlingen, Germany). Numerous clots and a large hemothorax in the left thoracic cavity were observed. Two additional ports were then inserted under direct vision. After the entire thoracic cavity had been rinsed with saline and clots had been removed, pulmonary arteriovenous fistulas were visible on the surface of the lung (Figure 4). No active bleeding was observed, and no further operations were required. Two 28F chest drains were introduced through the trocar incisions, one in the eighth and one in the anterior fifth intercostal spaces. The tubes were connected to an underwater seal suction device (Pleurevac; Genzyme, Cergy-Pontoise, France) with a negative pressure of 20 cm H₂O.

The patient was extubated 3 hours after the operation, and pain was managed with a patient-controlled analgesia pump. Intercostal drainage was continued for 4 days, with a total volume of 975 mL. No air leak was observed, and the postoperative hospital stay was 6 days.

Follow-up angiography at 1 month and CT scanning at 3 months revealed recurrent limited flow in the lingula fistula (Figure 5). Repeat embolization was performed with 2 coils (1 fibered coil of 2 × 50 mm and 1 coil of 2 × 30 mm) placed with a 0.018-inch microcatheter (Boston Scientific). The occlusion was completed without injection of embucilate. Follow-up CT scans at 3 months and 2 years showed no recurrence.

Discussion

Rupture of pulmonary arteriovenous malformations (PAVMs) is a rare life-threatening complication. Puskas and colleagues¹ did not observe any hemothorax in 21 patients treated between 1964 and 1992. Ference and coworkers² reported only 5 (3.5%) patients with spontaneous hemothorax and 6 with massive hemoptysis in a study of 143 patients with PAVMs and hereditary hemorrhagic telangiectasia.

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Received for publication March 13, 2003; accepted for publication April 24, 2003.

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J Thorac Cardiovasc Surg 2003;126:1204-7

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0022-5223/2003 \$30.00 + 0

doi:10.1016/S0022-5223(03)00806-7

Figure 1. Thoracic CT scan showing left hemothorax and lobulated opacity in the left lower lobe with uptake of contrast.

Figure 2. Pulmonary angiogram demonstrating PAVMs in the upper part of the left lower lobe (A) and in the lingula (B). The diameter of the segmental feeding artery of the lingula fistula was 5 mm.

Figure 3. Pulmonary angiogram after embolization and complete obliteration of PAVM.

In most reported cases of PAVMs, the routine approach is preventive transcatheter embolization to avoid complications. These include neurologic sequelae (ie, stroke, transient ischemic attack, cerebral abscess, and seizures) and, less commonly, pulmonary hemorrhage, which could be fatal.^{3,4}

In contrast to reported cases, our patient was treated in a life-threatening situation. The large hemothorax, as well as the previous medical history, prompted us to perform a CT scan to investigate the presence of PAVM. As reported by Martinez and associates,⁵ spontaneous hemothorax frequently occurs in patients with disease-related coagulopathy or those receiving anticoagulation treatment. Other causes include intrapleural bleeding compli-

cating pneumothorax (ie, disruption of pleural adhesions or torn vascular adhesions), tuberculosis, sarcoidosis, pulmonary infarction, ruptured thoracic aneurysm, and subdiaphragmatic pathology (endometriosis and pancreatic disease). The CT scan carried out on our patient confirmed the presence of PAVMs.

Because the patient's hemodynamic condition was stable, we decided to perform a transcatheter embolization despite the presence of abundant pleural clots and hemothorax.

In our case the arteries entering the PAVMs were technically suitable for coil occlusion, being long enough and not enlarged just before entry into the aneurysms, without significant risk of the coils entering the aneurysms and the draining veins. When feeding arteries are short (<3 cm), occlusion of the aneurysm might be

Figure 4. Thoracoscopic view showing the thrombosed PAVMs on the left pulmonary surface: **A**, PAVM located in the upper part of the left lower lobe; **B**, PAVM located in the lingula.

Figure 5. Follow-up thoracic CT scan 3 months after embolization, showing partial recanalization of the lingula fistula.

necessary.⁶ Detachable coils can be withdrawn if they are not correctly sized or can be reinserted if not properly placed. However, some authors prefer to use detachable balloons.³

Supplying arteries with a diameter of 5 to 9 mm (approximately 70% of PAVMs) can be occluded equally well with either detachable silicone balloons or coils. Personal experience and preference for a certain device will often be decisive in the choice of technique.⁷ Because anatomy can vary greatly, both detachable silicone balloons and coils should be available as therapeutic options.⁴ After deployment of several coils in the feeding artery, flow is reduced, and embolization can safely be achieved with glue, which can shorten the procedure.

In most cases PAVMs are multiple, and more than one embolotherapy session is required per patient. In a study reported by White,⁶ 276 PAVMs were diagnosed in 76 patients; all patients

had multiple malformations, and on average, 2 embolotherapy sessions were necessary per patient in addition to diagnostic angiography, which was performed on a separate day. Embolotherapy sessions were separated by 4 to 6 weeks.

Current indications for transcatheter embolization include the prevention of complications in larger (feeding artery diameter >3 mm) and symptomatic PAVMs. From our experience in cases of massive hemoptysis-hemothorax, we believe occlusion of all existing PAVMs should be considered. This approach might have the additional benefit of reducing the risk of paradoxical emboli and other complications associated with unoccluded PAVMs.² To our knowledge, no data are currently available to predict which PAVMs is most likely to rupture on the basis of size or location.

After embolization, different therapeutic options were available. A thoracic drainage procedure without surgical intervention was an option; however, drainage efficiency could be hindered by the presence of numerous clots. A surgical procedure to remove clots and confirm the absence of bleeding was therefore considered essential. Two surgical strategies were available. The first strategy was a thoracotomy with local excision of all the PAVMs. This technique, described by Bosher and colleagues,⁸ can be combined with a previous embolization procedure.⁹ The feasibility of selective surgical resection depends on the PAVM localization, however, and lung tissue sparing is not always achievable. The second option was to perform video-assisted surgery with wedge resection of the PAVM, which could avoid the higher morbidity rates of thoracotomy. This technique has been previously described by Temes and associates¹⁰ but only for a solitary malformation. In our case the PAVMs were multiple and not easily accessible for video-assisted thoracoscopic resection. Because of a complete absence of PAVM bleeding, we decided only to remove blood clots.

As previously described by several authors,^{11,12} we observed a recanalization of a PAVM 3 months later. Usually, occluded PAVMs disappear or are reduced to a fibrous strand by the end of 1 year. Any evidence of persistence on CT suggests recanalization (16% in the study of Ference and coworkers²) and is an indication for re-embolization.³ Thereafter, follow-up thoracic CT scanning is recommended every 3 to 5 years to investigate the possible development of new PAVMs or growth of small PAVMs.³ Some authors have proposed the use of 2-dimensional contrast echocardiography, with good results in detection and follow-up.^{13,14}

All patients with hereditary hemorrhagic telangiectasia should undergo routine screening for PAVM with noninvasive techniques, preferably with the 100% oxygen method. Family members of patients with hereditary hemorrhagic telangiectasia should also be screened for PAVM.¹⁵

In our case a medical history of hereditary and recurrent epistaxis, as well as the presence of lip telangiectasia, was sufficient to suspect ROW syndrome and screen for PAVMs, which are associated with ROW in 60% to 90% of cases.¹⁶

Conclusion

Even in the presence of PAVM rupture, transcatheter embolization could be performed. This procedure, combined with video-assisted thoracoscopy, avoided lung tissue loss, preserved lung function, and minimized morbidity. Because PAVMs can be recurrent, minimally invasive treatment options are essential. Although re-

peated procedures might be required for persistent PAVMs or recanalization, transcatheter embolization is a lung-sparing procedure and should therefore always be considered in cases of PAVM.

We thank Mr Richard Medeiros and Miles Dalby for their advice in editing the manuscript.

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Fatal air embolism during computed tomography-guided pulmonary marking with a hook-type marker

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Pulmonary marking with a hook wire, guided by computed tomography (CT), is often used to assist in the resection of small lung tumors that are difficult to identify by means of thoroscopic surgical techniques or digital palpation. The most common complications accompanying

this procedure are pneumothorax, intrapulmonary hemorrhage, and hemothysis. These complications are usually mild, self-limiting, and curable. Air embolism is a widely recognized complication of transthoracic percutaneous needle biopsy, but in the case of CT-guided pulmonary marking, few complications have been reported. We report a case of fatal arterial gas embolism during hook wire localization of a pulmonary lesion. The patient mortality was mainly attributed to ischemic heart injury caused by air embolism. Physicians should be aware of this possible complication when a hook wire-type marker is used to avoid further fatalities.

Clinical Summary

A 79-year-old woman visited our hospital for an examination of a 2.0-cm abnormal shadow in the left upper lung field revealed by a mass chest radiographic survey. A chest CT demonstrated a 1.5-cm tumor shadow with a faint density and ground-glass opacity in S⁸ of the right lung in addition to a 2.0-cm tumor in S¹⁺² of the left lung (Figure 1).

The patient was known to have had pulmonary tuberculosis in her late twenties. She had no smoking history. Results of her

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Received for publication March 21, 2003; accepted for publication April 24, 2003.

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J Thorac Cardiovasc Surg 2003;126:1207-9

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0022-5223/2003 \$30.00 + 0

doi:10.1016/S0022-5223(03)00821-3