DETACHABLE COILING FOR EMBOLOTHERAPY OF HIGH-FLOW PULMONARY ARTERIOVENOUS MALFORMATION: CASE REPORT AND REVIEW OF THE LITERATURE

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Pulmonary arteriovenous malformations (PAVMs) are very rare anomalies of the lung parenchyma. Although many patients are asymptomatic, it carries a high risk of severe cerebral complications, and rarely fatal outcome due to rupture of the malformation.
There is a strong association with the Rendu-Osler-Weber disease.
The patients should be treated in case of symptomatic PAVMs (paradoxical emboli), and also preventively, when the feeding artery is at least 3 mm in diameter.
Embolization is a well-established treatment, which can be very challenging when dealing with very high-flow fistulas, where there is a risk of inadvertent migration of the embolization material in the left cavities of the heart and in the aorta.
This report emphasizes the importance of the diagnosis and the embolization indication in pulmonary arteriovenous malformations, and the usefulness of detachable coils in case of high-flow fistulas.

Key-words: Arteriovenous malformations – Embolism, pulmonary.

Case report

A 72-year-old man was treated for sigmoid adenocarcinoma, staged T2N0. A routine follow up chest radiograph, showed a round nodular lesion in the right lower-lobe (Fig. 1).

Chest Angio-CT scans demonstrated an artery-to-vein pulmonary fistula, with a very large associated aneurysm (Fig. 2). No cardiac or cerebral complications were present.

The diameter of the arterial feeder (6.6 mm) and of the aneurysm indicated the need for a treatment.

Embolization was performed under general anesthesia, by femoral vein puncture and catheterization of the right lower pulmonary artery. Then, an exchange for a 90 cm long 5 French flexible introducer sheath (Flexor Shuttle-SL®, William Cook Europe Aps, Bjaeverskov, Denmark) was performed over a extra stiff guide wire (Amplatz TFE coated®, William Cook Europe Aps, Bjaeverskov, Denmark), in order to obtain a stable position.

A 4 French catheter (vertebral, Optitorque®, Terumo Europe NV, Leuven, Belgium) was then advanced as close as possible to the origin of the aneurysm.

A long detachable 0.035-inch coil of 12 mm diameter and 6 cm long (Jackson Detachable Embolization Coils Inconel-Mreye®, William Cook Europe Aps, Bjaeverskov, Denmark)

Fig. 1. — Chest X-Ray shows a vascular mass in the right lower lobe (arrow).

was then progressively pushed, until a safe and stable position could be obtained, before delivery (Fig. 3).

Several 0.035-inch coils (one Nester® of 12 mm diameter and 14 cm long and two Nester® of 10 mm diameter and 14 cm long, Embolization Coils, and two Embolization Coils Inconel-Mreye®, William Cook Europe Aps, Bjaeverskov, Denmark) completed the occlusion (Fig. 4).

The control chest radiograph performed 6 months later showed no dislocation of the embolization material, and the control thoracic angio CT scan confirms good occlusion of the pulmonary arteriovenous shunt (Fig. 5).

There was no complication related to the procedure.

Discussion

A PAVM presents as an abnormal communication between the arterial pulmonary tree and pulmonary veins. It may be simple (one feeding artery, one outflow vein) or complex (multiple feeders, potentially intercostals or bronchic (1, 2).

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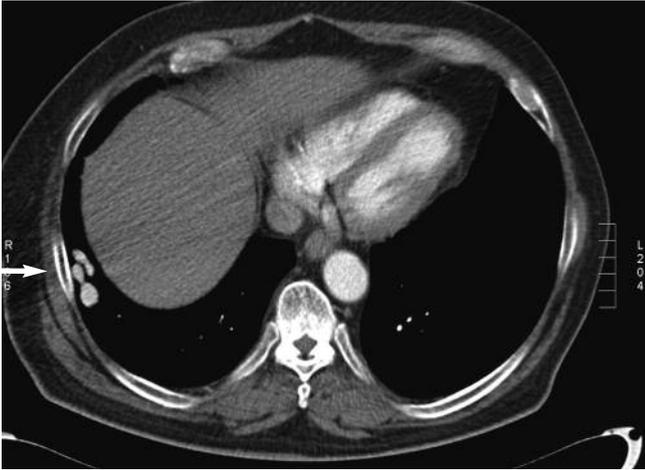E-mail: marc.laureys@chu-brugmann.be

Fig. 2. — On angio CT scan, there is an intensely-enhanced pulmonary artery to vein malformation (arrow).
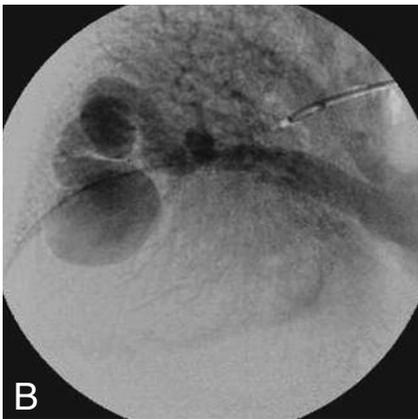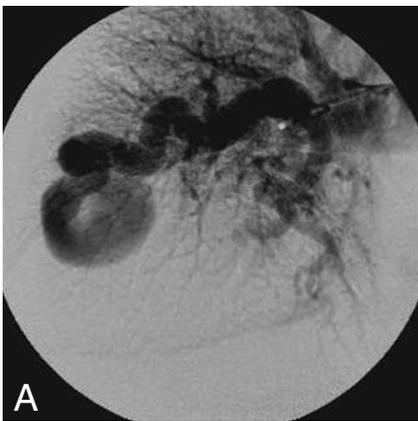

Fig. 3. — A-C. The selective pulmonary angiography depicts the malformation. We see the implantation of the first detachable coil followed by several Nester® coils (C).

A thin-walled aneurysm, or pseudoaneurysm, located on the venous side, is often associated (3).

60 to 90% of pulmonary arterio-venous malformations (PAVMs) are associated with Hereditary Haemorrhagic Telengectasia, also known as Rendu-Osler-Weber disease (4).

Acquired causes of PAVMs include post-thoracic surgery or trauma, infections (tuberculosis, actinomycosis, schistosomiasis), long-standing cirrhosis, metastatic carcinoma, mitral stenosis, and systemic amyloidosis (1, 5).

PAVMs are a potential source of complications, related to paradoxical embolism. It is due to the absence of a normal filtering capillary bed, that may led to serious cerebral sequelae in the cerebral circulation: these include stroke (18%), transient ischemic attacks (37%), cerebral abscess (9%), migraine (43%) and seizures (8%) (1, 6).

Rarely, the abnormal vessels may rupture into the pleura, the bronchus, or the pulmonary parenchyma (7).

Diagnosis

Chest radiography may show the classic feature of PAVM: a round or lobulated lesion, varying in size from 1 to 5 cm in diameter, generally located in the lower lobes (1).

Contrast echocardiography (with intravenous injection of agitated saline to create microbubbles) is useful to establish the diagnosis of right-to-left shunt, but is inaccurate in quantifying the shunt fraction, which is done by radionuclide imaging, or far less costly, by the 100% Oxygen method (8).

Angio CT scans (Multirow Detector Computed Tomography, with intravenous injection of iodinated contrast material) allows a high detection rate of PAVMs, especially when multiple. Remy et al. (9) compared pulmonary angiography with Angio CT and found significantly better detection of PAVM with Angio
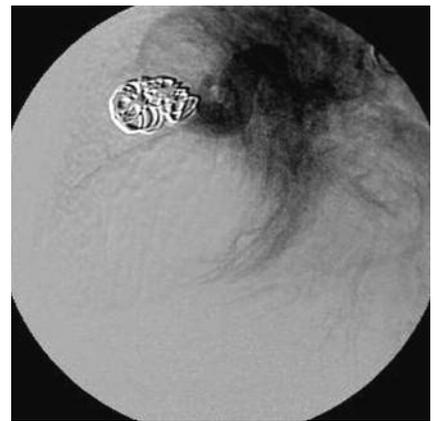

Fig. 4. — The control angiograms shows the total occlusion of the PAVM.
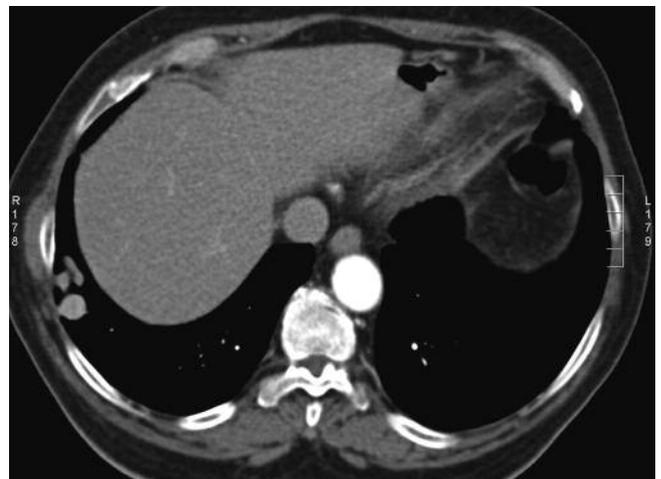

Fig. 5. — Control angio CT scan at the same levels, with no residual enhancement of the fistula.

CT (98% versus 60% with pulmonary angiography).

If a surgical or endovascular treatment is planned, digitally subtracted pulmonary angiography is required to obtain detailed information about the angioarchitecture, the morphology and the complexity of the PAVM (10).

Treatment

All symptomatic PAVMs must be treated, even when small, as morbidity in untreatred patients is high (50%) compared to the treated patients (3%) (11).

When the feeding artery is greater than 3 mm in diameter, embolization is indicated in order to prevent the patient from complications (strokes and brain abscess) (12).

Portsmann first reported embolotherapy for PAVM (13). Embolization can be performed with various materials, but polyester fibers platinum coils are generally used.

It is mandatory to perform all exchanges "underwater", to prevent air from going through the PAVM during the procedure.

In high-flow fistulas, a detachable balloon may be implanted, but this latter is very expensive.

Inflation of an occlusive balloon is an alternative; it allows the deployment of the coils and prevent their migration (3). However, it is not always applicable and is rather unsafe as a migration risk still exists during the balloon deflation and retrieval.

More recently, detachable coils have been available. The device helps the interventional radiologist to deploy the first "security" loops progressively and safely, without threatening from the rapid flow. The deployment is done as close as possible to either the site of the fistula, or the aneurysmal sac. Other "conventional" coils can then be pushed into this nest, in order to achieve the occlusion.

The first successful surgical resection of a PAVM was reported in 1942 (14).

Nowadays, lung conservative surgery and thoracoscopic assisted techniques are described, that however carry the complication risk of all other thoracic surgery procedures. Disadvantages also include the pos-

sible loss of pulmonary parenchyma surrounding the PAVM, and the longer hospital stay (3).

Surgical resection is thus indicated in cases of intrapleural rupture or lesions not amenable to embolotherapy (8), although up-to-date embolization technique and material lead to successful occlusion in the near-totality of the patients.

Complications

They are encountered in less than 10% of the patients, and include localized and short-lasting pleurisy, pulmonary infarction distal to the occlusion, sepsis, and retrograde pulmonary embolism in case of polycythaemia (12).

Results

Immediate closure is achieved in 98% of the cases (12).

The long term follow-up shows a recanalization potential of the embolized site in about 8% of all the PAVMs (3, 15), and is not related to the embolization material (balloons or coils). Several studies show recanalization rates varying from about 3% to more than 20% (3), and is significantly higher when multiple PAVMs are present, and when all the visible PAVMs are not embolized (3).

Thus, new PAVMs may appear during the follow-up period, requiring new possible embolizations.

The lack of clinical response to occlusive or surgical therapy is related to the severity of the pulmonary disease (multiple PAVMs) (3).

Conclusion

Even when they are treated, they must be watched closely with thoracic angio-CT scans, as recanalization or new PAVMs may occur.

Embolotherapy remains a safe and efficient therapeutic method, carrying a very low complication rate, and has many advantages over surgery.

If the PAVM is a high-flow lesion, embolization can be performed with detachable coils, which allow a safe and stable first nest deployment.

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