Percutaneous embolization of pulmonary arteriovenous fistulas

Embolização percutânea de fistulas arteriovenosas pulmonares

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ABSTRACT - Pulmonary arteriovenous malformation is an abnormal communication between an artery and a vein, causing clinical manifestations, such as chronic hypoxemia and embolic events. Arteriovenous malformations were treated surgically, with a significant rate of complications. In the 1970's, the first percutaneous catheter embolization was performed with coils. We describe three cases in which percutaneous embolization techniques were effective to prevent embolic, hemorrhagic, and hypoxemic events in the follow-up of patients.

Keywords: Embolization, therapeutic; Arteriovenous fistula; Arteriovenous malformations; Pulmonary artery

RESUMO - Malformações arteriovenosas pulmonares são uma comunicação anormal entre uma artéria e uma veia, causando manifestações clínicas, como hipoxemia crônica e eventos embólicos. As malformações arteriovenosas eram tratadas cirurgicamente, com taxa expressiva de complicações. Na década de 1970, a primeira embolização percutânea por cateter foi realizada com molas. Descrevemos três casos nos quais técnicas de embolização percutânea foram efetivas em prevenir eventos embólicos, hemorrágicos e hipoxêmicos no seguimento dos pacientes.

Descritores: Embolização terapêutica; Fístula arteriovenosa; Malformações arteriovenosas; Artéria pulmonar

INTRODUCTION

Arteriovenous malformations (AVMs) are abnormal communications between a vein and an artery that can lead to clinical manifestations, such as chronic hypoxemia or embolic events, due to loss of capillary barrier, allowing the passage of thrombi and greater susceptibility to infections, since this erroneous communication also favors the passage of microorganisms directly into the arterial circulation.\textsuperscript{1} Approximately 80\% of cases of pulmonary AVMs (pAVM) are associated with hereditary hemorrhagic telangiectasia, an autosomal dominant disease also known as Rendu-Osler-Weber syndrome. Other acquired conditions are rarer, such as liver cirrhosis, mitral stenosis, Fanconi syndrome, metastatic thyroid cancer, and other cancers, in addition to idiopathic causes.\textsuperscript{2} The incidence of pAVM in patients with hemorrhagic telangiectasia is approximately 30 to 40\% per year; about 85\% of them are simple, in which one or more arteries originating from a single lung segment supply the malformation. Complex lesions have arterial feeding vessels from more than one lung segment and account for 5 to 10\% of cases.\textsuperscript{3} Interventional cardiology has experienced extraordinary progress. In the past, pAVMs were surgically treated, with a significant rate of complications and an increase in mortality rate.\textsuperscript{4} In 1977, Porstmann et al. performed the first catheter embolization using coils, opening up the possibility of percutaneous treatment.\textsuperscript{5} Approximately 57\% of patients are asymptomatic.\textsuperscript{6} The most common signs and symptoms are fatigue, exertional dyspnea, palpitations, cyanosis and hemoptyis, dyspnea.
being the most frequent.\(^7\) Percutaneous occlusion is indicated in all symptomatic patients (cyanosis, hemoptysis and recurrent strokes) and in all those who have fistulas with arteries >3mm in diameter, who are at greater risk of paradoxical embolism.\(^7\)

In this article, we report three cases of arteriovenous fistulas with varied clinical features, descriptions of the procedures used, and the results obtained. The Research Ethics Committee of the Hospital de Urgências de Goiânia, associated with the Plataforma Brasil, approved the present study (CAAE: 85497418.2.0000.0033).

**CASE REPORTS**

**Case report 1**

A 47-year-old male patient was referred to the hospital for evaluation of an arteriovenous fistula found in an elective chest tomography examination, which showed a pAVM characterized by a right pulmonary artery measuring 8mm in diameter, with a fistular sac in the subpleural topography measuring 4x18x33mm, drained by a subsegmental vein into the right inferior pulmonary vein (Figure 1A). The patient was asymptomatic from the pulmonary point of view. A right femoral vein puncture was performed under local anesthesia, and a 7F introducer was positioned, and a 5F JR guide catheter and a 0.035”x180cm guidewire were passed up to the branch of the pulmonary artery that supplied the fistula on the left. A 0.018x300mm guidewire and microcatheter were passed up to the fistula nidus, and a new angiography was performed to ensure correct positioning. Microcoils of 2.5x50cm (largest diameter) to 1.4x30cm (smallest diameter) were sequentially deployed. A post-intervention angiography confirmed the success of the procedure and complete exclusion of the fistula. Complete embolization was performed using 11 microcoils, with no complications or residual shunt. (Figure 1B). During the postoperative follow-up, the patient remained asymptomatic and with no complications in the 12 month-period after the procedure.

**Case report 2**

A 48-year-old female patient with rheumatoid arthritis and dyslipidemia as comorbidities, was diagnosed as pAVM a few years ago, associated with chronic hypoxemia, and was under home oxygen therapy, with an indication for fistula embolization. The patient was admitted to the emergency room with severe hypoxemia. A previous angiography showed an arteriovenous fistula fed by several branches of the right pulmonary artery (Figure 2A). Embolization of four lower sub-branches of the right branch of the pulmonary artery was performed using four vascular plugs (Figure 2B), with immediate improvement in oxygen venous central saturation (76% to >92% in room air). Embolization with Amplatzer® vascular plugs was chosen. A JR 5F guide catheter and a 0.035”x300cm Amplatzer Super Stiff™ guidewire were passed up to the right branch of the pulmonary artery. A pre-intervention angiography was performed, and four vascular plugs with the following diameters, AVP II 6x6mm, AVP II 14x10mm, AVP II 3x6mm, and AVP II 6x6mm, were deployed, under angiography, and successfully positioned in the four fistulas, achieving total exclusion of the vessel. There were no complications in the 12-month postoperative follow-up.

**Case report 3**

A 30-year-old male patient had an atrial septal defect corrected in 2007 as comorbidity. A coronary cineangiography with pulmonary arteriography was performed due to an accidental finding of a nodular image on a chest CT

![Figure 1](image1.png) The arteriovenous fistula before (A) and after embolization (B).
scan. During the examination, an important aneurysm was observed, originating in the artery of the left upper lobe branch, measuring 21.47x24.86mm. The walls were calcified and showed turbulent and slow flow inside (Figure 3A). Two important AVMs were observed in arteries of the right lower lobe, resulting in an important right-to-left shunt (Figures 3B1 and 3B2). Percutaneous embolization of the fistula of the superior branch of the right inferior lobar artery of 12mm was performed with 14 microcoils and the fistula of the middle branch of the right inferior lobar artery of 13mm with 9 microcoils and the fistula of the inferior branch of the right inferior lobar artery was occluded with a plug vascular AVP II 6mm (Figure 3C). The aneurysm was occluded using 16 microcoils, and a 5mmx19mm stent was implanted in the left superior lobar artery (Figure 3D). The embolization of the fistulas was effective, and there was no recurrence or complications in the 12-month follow-up.

Figure 2. The arteriovenous fistula before (A) and after embolization (B).

Figure 3. Aneurysm (A). Arteriovenous fistulas (B1 and 3B2); after embolization of arteriovenous fistulas (3C); after occlusion of the aneurysm (D).
DISCUSSION

In these three clinical reports, we observed two simple fistulas originating in the lower right and lower left lobes, and one complex fistula associated with an aneurysm. Percutaneous embolization was successfully performed in all contexts, both in the simple cases, using microcoils, and in the complex one, using vascular plugs, with no need for a new approach to correct complications in 12 months of follow-up. The results reproduce those observed in other studies.\(^\text{8,9}\)

The adequate and precise choice of material to be used for the percutaneous closure is important to reduce procedure failure and complications. Treatment options include percutaneous embolization with coils, a detachable balloon, or other devices, as well as surgical excision. The coils must have a diameter 2mm larger and the plugs must be 20 to 30% larger than the vessel to be occluded. Untreated lesions are associated with an 11% mortality rate and a 26% morbidity rate.\(^\text{10}\)

In the first and third reported cases, embolization with microcoils was performed in the context of avoiding paradoxical embolism based on the diameter of the pulmonary arteriovenous fistula. Both patients were asymptomatic. Currently, there is no randomized study comparing the better choice between clinical and surgical treatments of fistulas. However, there is evidence that percutaneous treatment with occlusion reduces the probability of cerebral embolization, and this treatment is indicated in all symptomatic patients (cyanosis, dyspnea and paroxysmal embolism) and in those who present with a fistular sac >3mm in diameter, due to a higher incidence of embolic events.\(^\text{8}\)

In cases with large caliber fistulas, in which the use of coils is not feasible due to the chance of their embolization and important complications, the use of vascular plugs, associated or not with coils, is the best approach, as also described in the case reports by Erudilho et al.\(^\text{8}\) and Faria et al.\(^\text{9}\)

The surgical approach is restricted to complex cases, when the supplying arteries have very large calibers, and the vascular anatomy does not favor percutaneous intervention, or cases with unsuccessful embolization and/or that have not achieved satisfactory results.\(^\text{11}\)

In the three cases, a surgical approach was not necessary after the percutaneous procedure. In the series of cases presented by Salibe-Filho et al., only one patient required corrective surgery after unsuccessful embolization.\(^\text{4}\) Surgical treatment includes lobectomy, resection or ligation of the fistulas.\(^\text{12}\) Lung transplantation is an exceptional measure.\(^\text{5}\) The treatment of multiple fistulas can be performed in a single procedure, as in the series here described; however, in some individuals with a large number of AVMs or when several of them are complex, the best option is to perform several embolization sessions distributed on different days, as we did in our third case reported.

The main complications of percutaneous embolization are pleuritic pain, pleural effusion (10 to 35%), infection, pulmonary infarction, and device embolization.\(^\text{8}\) Of the three case reports, none of the patients presented complications after the procedure.

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None.

CONFLICTS OF INTEREST

The authors declare there are no conflicts of interest.

CONTRIBUTION OF AUTHORS

Conception and design of the study: LXAO, AGA, MLP and GG; data collections: LXAO and DMF; data interpretation: LXAO, FACC and AGA; text writing: LXAO and GG; approval of the final version to be published: LXAO, FACC, DMF, MLP, AGA and GG.

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