Transcatheter Microcoil Embolization of an Idiopathic Solitary Pulmonary Artery Aneurysm

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Abstract

An incidental vascular abnormality was identified in a female patient in her 70s with a history of malignant lymphoma. Contrast-enhanced computed tomography (CT) scans revealed a pulmonary artery aneurysm (PAA) originating at the A10 branch of the right pulmonary artery (10 × 9 × 9 mm in size), which involved three distal branches at the aneurysmal sac. Retrospectively, this PAA was identified on CT images two years prior, and has been growing since. Endovascular embolization was performed with microcoils that were placed in the aneurysmal sac and the distal and proximal sites of the parent artery, achieving complete resolution of the PAA without complications. Coil embolization can be one of the treatment options in management of the unruptured PAA, although further investigations are necessary.

Key words: Pulmonary artery aneurysm, Peripheral, Solitary, Idiopathic, Transcatheter arterial embolization

Introduction

Pulmonary artery aneurysms (PAAs) are uncommon, representing 0.5-1% of all thoracic aneurysms [1]. A solitary peripheral PAA presenting in a patient with no risk factors, such as local infection, inflammation, or an iatrogenic condition, is extremely rare [2]. For this reason, there are no well-defined guidelines regarding treatment for PAA. In cases of PAA rupture, the resulting massive hemothysis may be fatal. Therefore, aggressive treatment such as coil embolization should be considered as an option for management of unruptured PAAs, especially for growing PAAs regardless of the etiology or the presence of any symptoms. The current treatments available for PAAs are surgery or transcatheter arterial embolization (TAE) [3]; both treatment modalities have their advantages and disadvantages. We report a case of idiopathic peripheral PAA and its treatment with TAE.

Case Report

The patient in this case is a woman in her 70s who had been diagnosed with malignant lymphoma of the base of the tongue several years prior to the current presentation. The lymphoma had been successfully treated with chemotherapy. Upon review of a follow-up fluorine-18-fluorodeoxyglucose positron emission tomography/computed tomography (18F-FDG PET/CT) scan, a suspected pulmonary arteriovenous fistula was noted. The patient was asymptomatic. Laboratory data revealed that the patient suffered from mild anemia. A contrast-enhanced chest CT revealed a PAA measuring 10 × 9 × 9 mm, originating at the A10 branch of the right pulmonary artery with three distal branches at the aneurysmal sac. (Figure 1). The diameter of the originating artery proximal to the aneurysmal sac was 4 mm. No abnormal findings were observed on the CT scan apart from the PAA. When compared with the CT image taken two years previ-
ously, it was noted that the aneurysm had increased in size by about 1 mm in each diameter. The patient was informed that PAAs are rare and that there are no fixed treatment guidelines, but that the increase in the size of the PAA was of concern in her case. The patient agreed to undergo endovascular embolization rather than surgical treatment.

The right femoral vein was punctured and a 6 French (F) sheath was inserted under local anesthesia; 3000 units of heparin were then intravenously injected from the sheath, and heparinization of the whole body was performed. Digital subtraction angiography (DSA) of the right pulmonary artery was performed using a 4F pigtail catheter, and the right A10 branch was selected using a 6F occlusion catheter (balloon diameter: 10 mm) (Patlive®, Terumo Clinical Supply, Gifu, Japan) to control the blood flow locally, prevent migration of the coils, and give adequate support to the microcatheter. The presence of the PAA was confirmed at this stage (Figure 2).

The distal branches of the aneurysm were selected using a microcatheter (Progreat™ B3, Terumo, Tokyo, Japan) and all three were embolized, using a total of 18 microcoils (Orbit Galaxy®, Codman Neuroendovascular, Johnson and Johnson, Raynham MA, USA: four measured 4 mm × 12 cm; one measured 4 mm × 10 cm; three measured 3.5 mm × 9 cm; one measured 3.5 mm × 7.5 cm; two measured 2.5 mm × 5 cm; two measured 2.5 mm × 2.5 cm; five measured 2 mm × 2 cm) (Figure 3). Subsequently, the microcatheter was inserted into the PAA itself and coil embolization of the aneurysmal sac and of its feeding artery was performed using five AZUR® CX microcoils (Terumo: one measured 10 mm × 32 cm; one measured 9 mm × 28 cm; one measured 8 mm × 28 cm; two measured 6 mm × 20 cm) and 6 Orbit Galaxy microcoils (two measured 4 mm × 8 cm; two measured 3 mm × 6 cm; one measured 3 mm × 4 cm; one measured 2.5 mm × 2.5 cm).

The volume of the PAA was calculated using the following formula, assuming that the PAA was elliptical:

Aneurysm volume = 4π (height/2 × length/2 × width/2)
Coil volumes were calculated using the following formula:
\[
\text{Coil volume} = \pi \text{ (radius)}^2 \times \text{ length}
\]
The coil packing density was calculated using the following formula:
\[
\text{Packing density} = \left( \frac{\text{coil volume}}{\text{aneurysm volume}} \right) \times 100\%
\]
The coil packing density of the aneurysmal sac was calculated to be 40.3%. A post-procedural DSA confirmed that blood flow to the PAA was completely occluded (Figure 4).

A follow-up plain CT scan carried out 6 months after the TAE indicated no coil migration or compaction. The patient reported no symptoms suggestive of complications.

**Discussion**

The incidence of PAA noted in one study was 0.007% (8/109571); the cases in this study were diagnosed during post-mortem examinations [1]. PAAs can be central (from the main trunk or the left and right main pulmonary artery) or peripheral (more distal). Peripheral PAAs are reported to represent 11% of all PAAs. Central PAAs often develop due to pulmonary arterial hypertension (PAH) following congenital heart disease; in contrast, peripheral PAAs are more often secondary to Behçet’s disease, a local infection, or trauma, including iatrogenic trauma [2]; hence, they are often found as pseudoaneurysms. Peripheral PAAs have a 40-60% chance of rupturing [4], and 54-87% of these cases of rupture are fatal [5]. Some researchers have suggested immediate treatment of PAAs regardless of their etiology, size, or the presence of symptoms [2, 4, 5]. Conversely, it has also been reported that PAAs occurring in the absence of PAH have a low risk of rupture, and conservative medical treatment is recommended [6]. In the current case, the abovementioned etiologies were excluded, and the patient was diagnosed as having an idiopathic PAA. The intervention in this case was determined due to an observed slight enlargement of the PAA; the patient’s request also determined the choice of treatment. There are still many uncertainties regarding the natural prognosis of “true” idiopathic peripheral PAAs. Although there are currently no definitive guidelines on the management of this type of aneurysm, Deb et al. reported that idiopathic PAAs are treated using the same hemodynamic forces as aortic aneurysms, and if cystic medial degeneration (CMD) is present, these aneurysms are prone to dissection and rupture [7]. It is recommended that patients with enlarged (> 6 cm) aneurysms that are central, as well as those of any size that are symptomatic, should receive treatment.

**Figure 3.** Embolization of three distal branches using 18 microcoils.

**Figure 4.** Aneurysm packing and embolization of the feeding artery using 11 microcoils. A: fluoroscopic image, B: DSA after embolization
Currently, the main treatments for PAAs are surgery and TAEs. The treatment modality must be selected according to the patient’s medical history (e.g., complications, respiratory function), patient’s request, and the lesion type (e.g., the size of aneurysm, whether it is central or peripheral). Although surgery has a high success rate, it has the disadvantage of being highly invasive. However, one of the advantages of TAE is the possibility of re-intervention for recanalization after long-term follow-up. Regarding the TAE treatment in this case, embolization of the feeding artery alone was judged as inadequate because the pulmonary artery could potentially have some communicating vessels, including the bronchial artery in some cases [8].

Owing to the concern regarding the possibility of retrograde blood flow from distal anastomoses with the bronchial artery tree, endovascular isolation and packing a large number of detachable coils was performed. In particular, the choice of “isolation and packing” instead of “isolation alone” was selected to make embolization as tight as possible for complete occlusion of sac. Since the possible fragility of the aneurysm wall could not be denied, the coil closest to the diameter of the aneurysm was used first for packing to reduce the pressure to the vessel wall. The coils thereafter were downsized. Although the treatment site was different, the goal of coil packing density was set to at least 24%, as Yasumoto et al. reported that no compaction or recanalization occurred in visceral aneurysms [9]. Hydrocoils were selected to reduce the incidence of recanalization with less reliance on thrombus formation. There are some reports of N-butyl-2-cyanoacrylate (NBCA) (Histoacryl®, B. Braun, Tuttingen, Germany) and self-expanding nitinol wire mesh (Amplatzer™ Vascular Plug, St. Paul MN, USA) as alternative embolic materials for PAAs [10], but these cases seemed to be limited to certain lesion sites and a range of vessel diameters.

This report has several limitations. First, the 6-month follow-up using CT did not provide a sufficiently long interval, and an inadequate follow-up imaging modality was used. More long-term follow-ups and enhanced MR angiography or conventional angiography are necessary for strict evaluation of recanalization of the aneurysmal sac. Second, this intervention was performed with no definite indication of treatment of true PAA.

TAE for idiopathic peripheral PAA is less invasive than surgical treatment, and is considered to be a useful treatment option. There is a need for discussion regarding the details of PAA embolization methodology in the future.

Conflict of interest: We have no conflict of interests to disclose.

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