Original Research

# Emergency Endovascular Management of Pulmonary Artery Aneurysms and Pseudoaneurysms for the Treatment of Massive Haemoptysis

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**Haemoptysis, pulmonary artery aneurysms, pulmonary artery pseudoaneurysms, embolizations, coils, covered stents.**

**Introduction:** In this study we report the results of endovascular techniques for the management of pulmonary artery aneurysms (PAA) and pseudoaneurysms (PAPS).

**Methods:** Cases with massive haemoptysis due to PAA and/or PAPS that were managed by endovascular means were included in the study. Clinical history and procedure details were analysed. Primary endpoints were immediate technical success and re-intervention rate; secondary endpoints were survival rate and relapse of bleeding.

**Results:** Among the 72 patients with massive haemoptysis who were treated in our department during an 8-year period, 6 patients with 6 lesions (2 PAA, 4 PAPS) fulfilled the inclusion criteria and were included in the study. The mean age was 46.5 years; mean lesion diameter was 12.7 mm (range 4-22 mm); underlying pathologies were vasculitis (n=2), erosion by necrotic tumour (n=1), previous lung surgery (n=1) and infectious disease (n=2). Four lesions were treated with coils, 1 with coils and a bare stent, and 1 with a covered stent. The technical success was 100%. Mean follow-up was 20.4 months. The re-intervention rate was 50%, but in only 16.6% was it related to the treated lesions. Survival rate was 66.6%. There were no major or minor complications.

**Conclusion:** Endovascular management offers a safe and effective solution for the emergency treatment of massive haemoptysis due to PAA and PAPS. A variety of endovascular devices may be used, according to the size and the anatomical location of the lesion.

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Massive haemoptysis is an emergency condition in which more than 300 ml of blood is expectorated from the bronchial tree within 24 h; it is associated with a mortality rate of over 50%.[1,2] In the vast majority of cases it is related to pathology of the bronchial arteries, while in less than 10% it results from pathology of the pulmonary artery.[1,3] In some cases, however, both systemic and pulmonary arteries can be the source of haemorrhage.

Pulmonary artery aneurysms (PAA) and pseudoaneurysms (PAPS) are relatively rare. Usually, they are related to vasculitis, especially Behçet disease and Hughes–Stovin syndrome, and to infectious diseases such as active tuberculosis (Rasmussen aneurysms), aspergillosis, and necrotising pneumonia, particularly in immunocompromised patients. They may also occur after vessel erosion by a necrotic tumour or from direct vessel injury.[4] Since treatment with open repair is accompanied by elevated morbidity and mortality, percutaneous endovascular techniques have been developed for their treatment.[5]

We report the technical and clinical results from 6 patients with massive haemoptysis secondary to PAA or PAPS who were managed with percutaneous endovascular techniques.

## Methods

Study approval was obtained from the Clinical Audit Department of our Hospital. The inclusion criteria were the following: 1) more than 300 ml of blood expectorated from the bronchial tree within 24 h; and 2) presence of PAA and/or PAPS. Emergency computed tomography (CT) angiography using a multidetector scanner was performed prior to any treatment, as a first-line investigational imaging modality for the identification of the bleeding source. Between April 2004 and April 2011, 72 patients with massive haemoptysis were investigated in our department with a view to endovascular treatment. In 6 cases PAA and or PAPS were identified in the initial CT scan and the patients were included in the study. In the rest of the cases endovascular treatment with embolisation of the bronchial arteries was performed.

The 6 patients who were included in the study were 4 men and 2 women with mean age 46.5 years (range 26-63 years). All patients were haemodynamically unstable prior to the procedure. A mean of 600 ml/24 hours (range 400-1200 ml) of blood was expectorated for the last four days prior to the procedure and the patients received a mean of 3 units of blood (range 2-7 units).

A total of 6 lesions (2 PAA and 4 PAPS) were identified on the CT scan. The mean lesion diameter was 12.7 mm (range 4-22 mm). The lesions detected were in the right lower lobe superior segmental artery (n=1), the distal branches of the right lower lobe posterior basal segmental artery (n=1), the left upper lobe posterior segmental artery (n=1), the lingular artery (n=1), the middle lobar artery (n=1), and the main stem of the left inferior lobar artery (n=1). The lesions were characterised as saccular (n=4) or fusiform (n=2).

The underlying pathology of the 2 PAA was Behçet's disease; there was 1 PAPS case due to erosion of a pulmonary artery branch from non-small cell lung carcinoma, 1 PAPS following left pulmonary lobectomy for the treatment of a small cell lung carcinoma, and 2 mycotic PAPS in immunocompromised intravenous drug abuse patients. The aneurysm types and aetiology are presented in Table 1.

### *Procedure*

Access from the right common femoral vein approach was used, with placement of an 11 cm, 7 Fr sheath (Avanti Sheath Introducer, Cordis, Ireland). A 5 Fr pigtail catheter (Tempo, Cordis, Ireland) was advanced to the bifurcation of the pulmonary artery through the inferior *vena cava* and the right atrium and an angiogram was performed, followed by selective catheterisation of the pulmonary artery branch with a hydrophilic guidewire (Radifocus, Terumo Europe) and a 4 Fr C3 catheter (Tempo 4, Cordis, Ireland). In the cases where the lesion appeared to have a relative narrow neck (3 lesions) the 4 Fr catheter was advanced into the aneurysmal sac and several 0.035" coils were deployed (Nester Embolisation coils, Cook Medical, Denmark). The sizes commonly used were 8, 10, and 12 mm diameter. Coils were pushed into the lesion until no further contrast filled the lesion (Figure 1). Another technique used was the "front and back door" (2 lesions), when the lesion appeared to have a wider neck but originated from a relatively small vessel; coils were deployed in positions proximal and distal to the site of origin of the aneurysm. In one lesion of the right lower lobe superior segmental artery, several coils were released into the aneurysmal sac and it was deemed necessary to deploy a bare stent (7 mm × 40 mm, Zilver Vascular Stent, Cook Medical, Denmark) in the segmental artery in order to avoid coil migration ("caging technique") (Figure 2). Finally, in one lesion of a wide-neck saccular pseudoaneurysm of the main stem of the left inferior lobar artery, a balloon-expandable covered stent (7 mm × 38 mm Advanta V12, Atrium Medical Corporation, NH, USA) was deployed across the aneurysm's neck (Figure 3). After the completion of the procedure a final angiogram was performed to evaluate the final result and demonstrate the occlusion of the lesion. Bronchial arteriography was not performed in the first instance in any of the cases.

### *Endpoints and definitions*

Technical success was defined as the angiographic exclusion of the aneurysm from the pulmonary circulation on the final selective angiogram and the immediate cessation of haemoptysis. Re-intervention was defined as the need for a further procedure due to relapse of bleeding. Re-intervention was characterised as related to the treated lesion if contrast filling of the lesion was seen in the control CT scan, or as non-

**Table 1.** Characteristics of the treated patients.

| Pt | Anatomy | Type and size | Underlying disease | Embolisation technique | Technical success | Complications | Re-intervention | Follow up |
|---|---|---|---|---|---|---|---|---|
| 1 | Right lower lobe superior segmental artery | 14 mm PAA | Behçet's disease | Coil packing and "caging technique" with a 7 × 40 mm self-expandable bare stent | Yes | None | Yes, new lesion detected in the right upper lobe apical segmental artery. Treated with new embolisation with coils | 78 months - well |
| 2 | Right lower lobe distal branches of posterior basal segmental | 10 mm PAPS | Mycotic | Coil packing | Yes | None | No | 6 months - well |
| 3 | Left upper lobe posterior segmental | 7 mm PAPS | Iatrogenic after left upper lobectomy for small cell carcinoma | Coil packing | Yes | None | Yes, small existing component in the previously treated lesion. Embolised again with new coils | Died after 10 months from tumour recurrence |
| 4 | Lingular artery | 4 mm PAPS | Mycotic | Coil packing | Yes | None | No | 1 months - lost |
| 5 | Middle lobar artery | 15 mm PAA | Behçet's disease | Coil packing | Yes | None | Yes, bronchial artery embolisation | 6 months - well |
| 6 | Main stem of the left inferior lobar artery | 22 mm PAPS | Erosion from non-small cell lung carcinoma | 7 × 38 mm balloon expandable covered stent | Yes | None | No | Died after 6 months from tumour recurrence |

related otherwise. Procedural-related complications were classified as major or minor according to international reporting standards.[6]

## Results

The technical success rate was 100%, with successful exclusion of the lesion from the pulmonary circulation in the final angiographic check and immediate cessation of haemoptysis. Coil migration did not occur in any of the cases; all patients became haemodynamically stable immediately after treatment and further transfusion was not deemed necessary in any case. All patients were discharged after 2-3 days. Mean follow up was 20.4 months (range 1-78 months). One patient was lost to follow up after 1 month.

Relapse of haemoptysis occurred in three patients (50%). In two of them the presentation was with another episode of massive haemoptysis. Both patients were affected by Behçet's disease. The CT angiogram performed on their admission confirmed that there was no enhancement in the previously embolised lesion (bleeding unrelated to the treated lesion). In the first patient, a new pulmonary aneurysm was identified in the right upper lobe apical segmental artery. Selective catheterisation and treatment with coils followed in that new lesion. The patient became stable and was followed up for 24 months without evidence of further recurrence.

In the second case, CT confirmed that the bleeding did not originate from the treated lesion in the middle lobar artery, nor from another lesion in the pulmonary arterial bed; however, the bronchial arteries appeared to be dilated in the CT picture. A bronchial artery angiogram

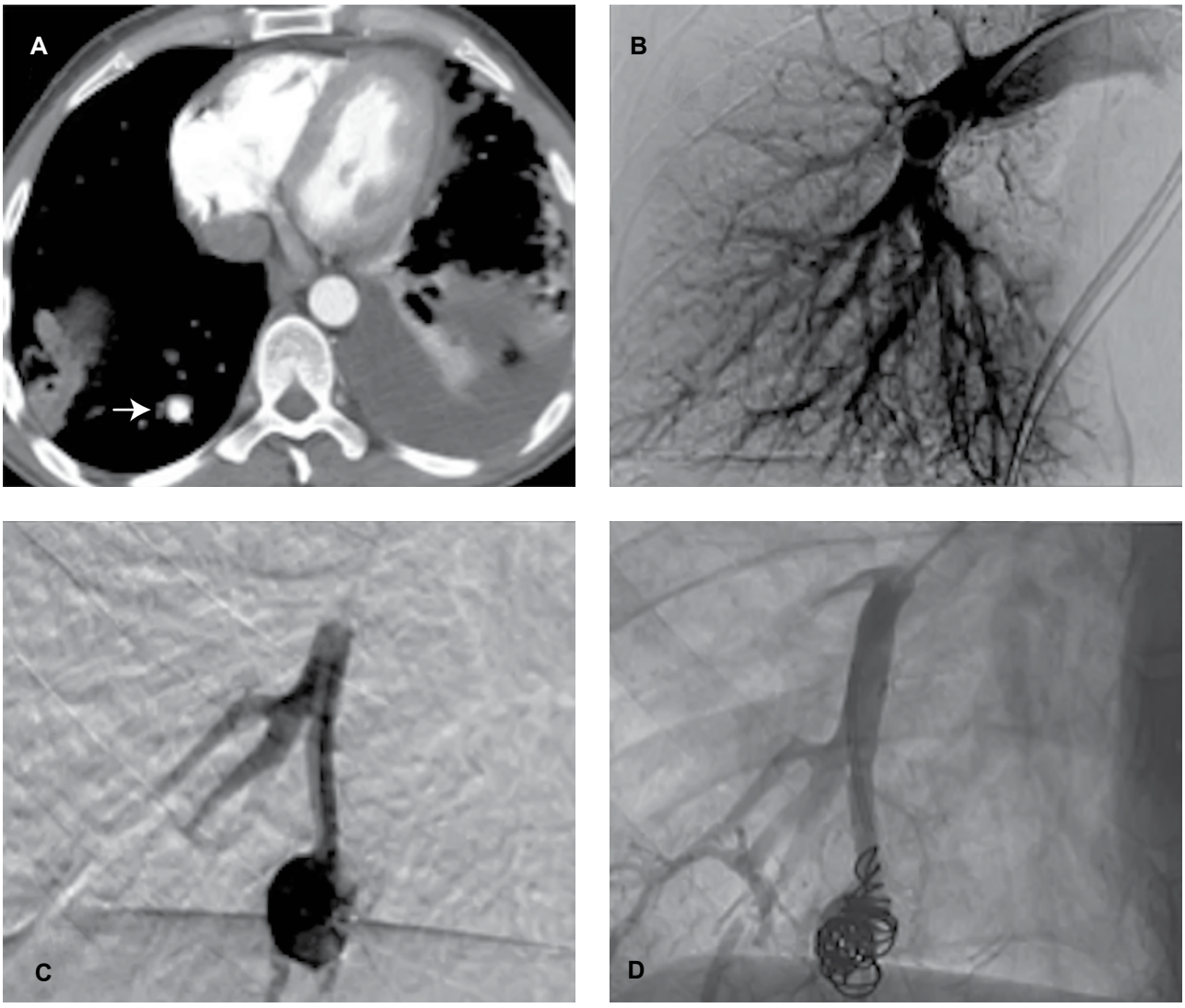


**Figure 1.** A. Computed tomography angiography demonstrating a 10 mm mycotic pseudoaneurysm of the distal branches of the right lower lobe posterior basal segmental artery (arrow). B. Pigtail angiogram confirms the presence of the lesion (arrow). C. Selective catheterisation of the right lower lobe posterior basal segmental artery with a 4 Fr catheter. D. Multiple coils deployed through the catheter into the aneurysmal sac.

was performed, with selective catheterisation of two arteries on the right and one on the left. The bronchial arteries appeared to be enlarged, with a beaded, pattern and were embolised with 300-500 μm polyvinyl alcohol particles (PVA, Cook Medical, Denmark). Haemoptysis stopped, and the patient became haemodynamically stable and was discharged two days later. There was no evidence of relapse after six months' follow up.

In the third case, intermittent haemoptysis occurred nine months after the initial embolisation of a pseudoaneurysm of the left upper lobe posterior segmental artery. The patient was readmitted, even though he was haemodynamically stable. A CT scan confirmed the presence of a small component adjacent to the previously embolised pseudoaneurysm. The lesion was successfully re-embolised using two additional coils. The patient died from other causes ten months later without recurrence of haemoptysis.

There were no major or minor procedure-related complications. Two patients died, both with lung malignancies, due to recurrence of the disease.

## Discussion

Pulmonary artery aneurysms and pseudo aneurysms are rare and their incidence is not known. They can be ei-

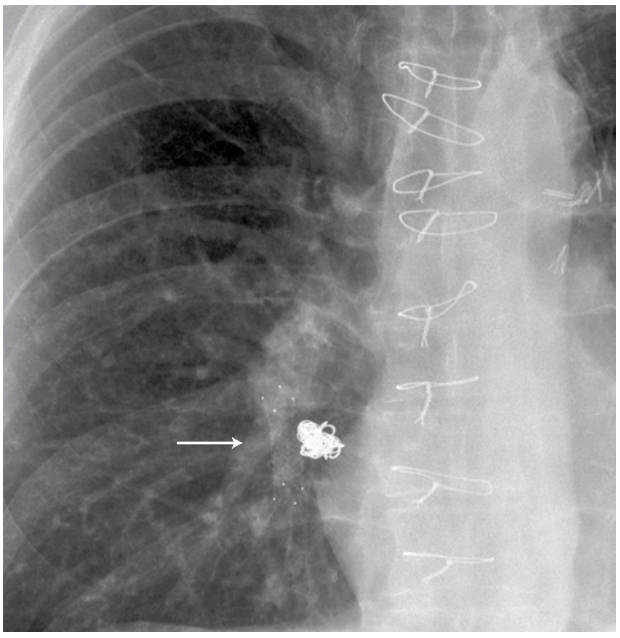

**Figure 2.** "Caging technique": a self-expandable bare stent (arrow) was deployed to avoid migration of the coils used for the exclusion of a 14 mm true aneurysm of the right lower lobe superior segmental artery.

ther idiopathic or caused by infection, systemic vasculitis, trauma, or erosion by a necrotic tumour.[4] The management options include traditional open surgical techniques (aneurysmectomy, lobectomy, graft replacement and patch repair) and percutaneous techniques.[5]

When the percutaneous route is chosen for the treatment of massive haemoptysis, there is always a question of whether to embolise the pulmonary or the systemic arteries. Our study has shown that, in the presence of PAA/PAPS, embolisation of the pulmonary arteries is enough for the immediate and midterm control of bleeding.

Other authors also agree that PAA/PAPS should be managed just by embolisation of the feeding pulmonary arteries, without embolisation of bronchial and non-bronchial systemic arteries. In a study by Remy et al,[5] among 189 patients treated for massive haemoptysis, in 11 the cause was attributed to pulmonary arterial origin. Five were shown angiographically to have a pulmonary arterial source of haemorrhage, one patient stopped bleeding only after embolisation of an apparently normal pulmonary artery segment, and five patients died of massive haemorrhage after embolisation of bronchial arteries only. The authors suggest that when a pulmonary arterial source of bleeding is suggested, the first thing to embolise is the pulmonary artery.

In another study by Sbano et al,[7] the same conclusion was drawn. Among 76 patients who underwent bronchial angiography and embolisation for haemoptysis over a 9-year period, 5 patients where identified in whom peripheral pulmonary artery pseudoaneurysms were found and embolisation of the aneurysmal pulmonary artery branch was performed. The authors conclude that embolisation of bronchial and non-bronchial systemic arteries alone may not be the indicated therapy to control haemoptysis in such cases, and occlusion of the pseudoaneurysm itself via a pulmonary artery approach is recommended.

The only suggestion for combined embolisation of pulmonary and bronchial arteries comes from Shin et al,[8] and only in the case of PAPS that originate from infectious processes and have a specific angiographic pattern. Combination bronchial embolisation is suggested only when the PAA/PAPS originate from an infectious process, there is suspicion of a bronchopulmonary arterial shunt, and the PAA/PAPS are too small to be seen on the pulmonary angiogram and are only seen in CT images. The bronchopulmonary arterial shunt may be revealed if PAA/PAPS are shown on the bronchial angiogram. However both situations are very rare. Double bleeding sources (systemic and pulmonary arteries) can also be suspected when the CT angiogram shows enlargement of the systemic arteries, in addition to the pulmonary artery pseudoaneurysm.[9]

In any of the above cases, the life-saving procedure is proved to be the direct treatment of the pulmonary aneurysm. There are several ways of treating a PAA/PAPS via an endovascular approach. In the literature there are some mini-series, such as the one from our department, and sporadic case reports. Most of the authors used coils[5,7,10,11] or microcoils.[8] Coils appear to be the most common solution for the treatment of PAA/PAPS because they offer the possibility of treating the lesion and preserving the originating pulmonary artery. The cost of conventional coils is rather low and in the cases treated no complications occurred, as in our series. The most worrying factor when deploying a coil is migration, particularly when the aneurysmal neck appears rather wide. However, in such cases the "caging technique" may be used, as in the case described in our series where a bare stent was used to protect the position of the coils.

Another solution may be the use of a plug. There is only one case in the literature involving treatment of a pseudoaneurysm of the left lower lobe pulmo-

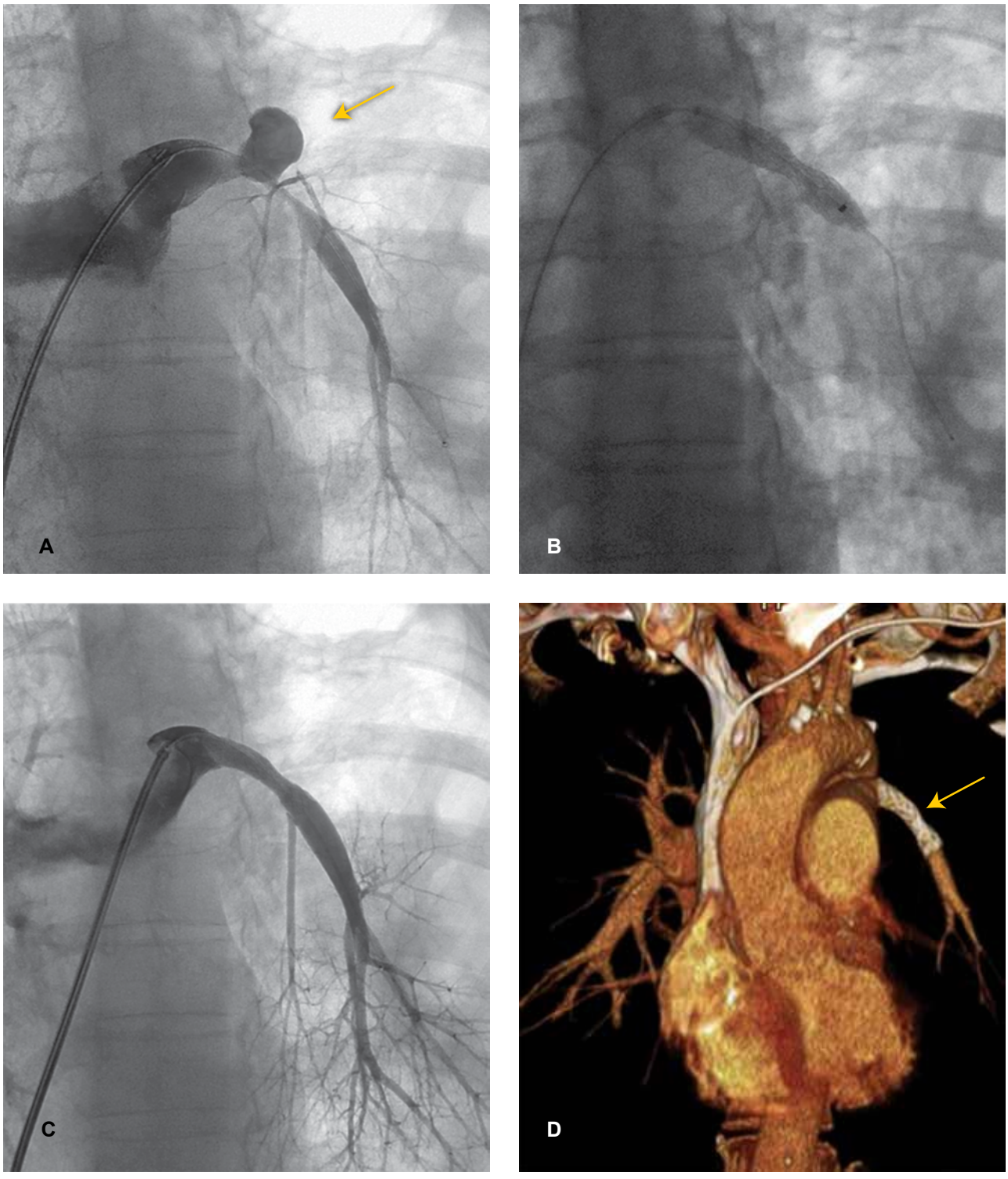


**Figure 3.** A. Selective digital subtraction angiography (DSA) demonstrating the presence of a 22 mm pseudoaneurysm of the main stem of the left inferior lobar artery (arrow). B. Fluoroscopic image demonstrating the deployment of a covered stent across the neck of the aneurysm. C. Final DSA confirming the complete exclusion of the pseudoaneurysm, as well as the preservation of the vessel's patency. D. Three-dimensional reconstruction of the multi-detector CT angiography demonstrating both lesion exclusion (arrow) and vessel patency, after 6 months' follow up.

nary artery with a patent *ductus arteriosus* closure device.[12] Even though the use of plugs may appear straightforward, there is a reason why coils are much preferred to plugs, particularly in the case of a pseudoaneurysm. When the pulmonary artery is irregular, e.g. when feeding a pseudoaneurysm in a necrotic tumour, a high radial force device like a plug may cause rupture due to the eroded arterial wall. In such cases, occlusion proximal and distal to the irregular pulmonary artery is considered to be the best practice.[7]

Some authors have also used liquid embolic materials for the treatment of PAA/PAPS, such as glue, ethylene vinyl alcohol copolymer, or thrombin.[13-15] However, even though there is no clear benefit from the use of liquid embolic agents, there is a risk of systemic artery embolisation and very serious complications. In addition, liquid embolic agents like ethylene vinyl alcohol copolymer also need to be prepared at least 15 minutes before use, and therefore are not suitable for an emergency setting. And of course, the cost of liquid embolic agents is much higher than that of conventional coils.

Finally there is a report of two cases treated with the use of a stent graft.[16] This might be a valid option in the case of a lesion with a very wide neck, as in the case treated in our series.

Our results demonstrated satisfactory management of a life-threatening condition in an acute setting. The minimally invasive nature of the procedure, the 100% technical success rate, the low long-term re-intervention rate, as well as the absence of procedural-related complications reported in this series demonstrate that endovascular procedures should be considered as a first-line treatment option in the management of massive haemoptysis caused by PAA and or PAPS.

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