Case Report

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Successful Endovascular Management of a Massive Hemoptysis due to a Rare Oncological Giant Pulmonary Artery Pseudoaneurysm

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Massive hemoptysis represents a distressing and life-threatening condition: 95% of cases originate from the bronchial artery system, with pulmonary arteries accounting for 6%–11% of cases. The most common etiology of hemoptysis from the pulmonary artery system is represented by pulmonary artery pseudoaneurysms (PAPs). PAPs are defined as the focal dilation of a pulmonary artery branch, involving only the adventice, with a higher risk of rupture than a true aneurysm. It constitutes a rare finding, often underdiagnosed by radiologists. However, PAPs can be life-threatening if undiagnosed due to a high mortality rate (50%). They frequently occur in patients affected by erosive inflammatory processes and necrotising infections of the lung or heart, but other etiological factors include trauma, vasculitis, neoplasia, pulmonary hypertension and Hughes–Stovin Syndrome. PAPs due to oncologic etiologies are rare. Among oncologic etiologies, the most frequent are represented by primary lung cancer rather than metastases. Today, CT angiography represents the imaging modality of choice; not only to establish diagnosis, but if performed with an appropriate timing of intravenous contrast, it also helps to plan therapy with an endovascular approach. In fact, endovascular treatment is the preferred therapeutic approach in managing hemoptysis due to PAPs, since a surgical approach is associated with a high risk of morbidity and mortality, especially in patients who are poor surgical candidates. In this article we report the case of massive hemoptysis due to an unusual giant PAP of the posterior lower branch of the right pulmonary artery, in a patient affected by pulmonary colliquated and confluent metastases, successfully treated by endovascular embolisation.

Keywords: *Hemoptysis; Pulmonary Artery Pseudoaneurysm; Endovascular Embolisation*

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INTRODUCTION

Pulmonary artery pseudoaneurysms (PAPs) represent an uncommon, but potentially fatal, cause of hemoptysis [1–3]. Their early identification is of fundamental importance, since massive hemoptysis from a ruptured PAP is fatal in more than 50% of patients [4]. PAPs are defined as a focal dilatation of a pulmonary artery branch, contained only by the outer vessel layer, constituted by the tunica adventitia [1,2]. They frequently occur in patients affected by erosive inflammatory processes and necrotising infections of the lung or heart, and in those who are at high risk of septic embolism [4,5]. Lung cavitations due to Mycobacterium tuberculosis infection have historically been associated with PAPs and these entities are known as Rasmussen aneurysms. Other etiological factors include trauma, vasculitis, neoplasia, pulmonary hypertension and Hughes–Stovin Syndrome [1,2]. Traumatic PAPs represent a very rare cause of hemoptysis and PAPs due to lung cancer are rare as well, with peripheral lung being, in oncologic patients, the most common site of

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onset [5–9]. In this article, we report the case of massive hemoptysis due to an unusual giant PAP of the posterior lower branch of right pulmonary artery in a patient affected by pulmonary colliquated metastases, successfully treated by endovascular embolisation.

Ethical Approval and Informed Consent

Ethical approval was not required. Informed consent was not required. The information has been anonymised.

CASE REPORT

A 68-year-old woman presented to our emergency department for repeated and worsening episodes of massive hemoptysis in the last 2 days, accompanied by severe dispnea. Her medical history was notable for mediastinal neuroendocrine carcinoma associated to pulmonary metastases, hypertension and type 2 diabetes mellitus; there was no history of smoking, trauma, fever, abdominal infections, or chest infections. Her general physical examination was unremarkable, she was not assuming anticoagulant therapy but her serum hemoglobin value was 5.9 g/dl. A contrast-enhanced CT scan of chest was performed, showing (Figure 1*a–d*) the lower right pulmonary lobe almost completely replaced by colliquated metastases, with evidence of a giant pseudoaneurym, with maximum transverse diameters of 30 × 28 mm, and a longitudinal diameter of 50 mm, of the posterior lower branch of the right pulmonary artery.

After a multidisciplinary discussion, the emergency surgeon, the vascular and thoracic surgeons and the interventional radiologist agreed to perform a selective angiographic study to better investigate the pseudoaneurysm and eventually perform a minimally invasive endovascular approach. In the angiographic suite, a selective right pulmonary artery angiography was performed, through a standard percutaneous venous right transfemoral approach, employing a 6 Fr long introductor-catether (80 cm Flexor Shuttle - SL Introducer, Cook Incorporated, 750 Daniels Way, Bloomington, IN 47404, USA), to get more stability of the coaxial system, and 5 Fr Cobra and MPA catheters (Cook Incorporated). Pulmonary artery angiography confirmed the CT finding, showing (Figure 2*a,b*) a giant pseudoaneurysm of the posterior lower branch of the right pulmonary artery. A super-selective catheterisation of the pseudoaneurysm's sac was performed employing a microcatheter (Progreat 2.7 Fr, Terumo, Shibuyau, Tokyo, Japan) followed by endovascular embolisation, through packing of multiple coils (POD, Penumbra, Inc. One Penumbra Place, Alameda, CA 94502, USA; Ruby Coil, Penumbra, Inc. One Penumbra Place, Alameda, CA 94502, USA) into the sac (Figure 3*a–c*) and its in-flow tract, finally sealing the

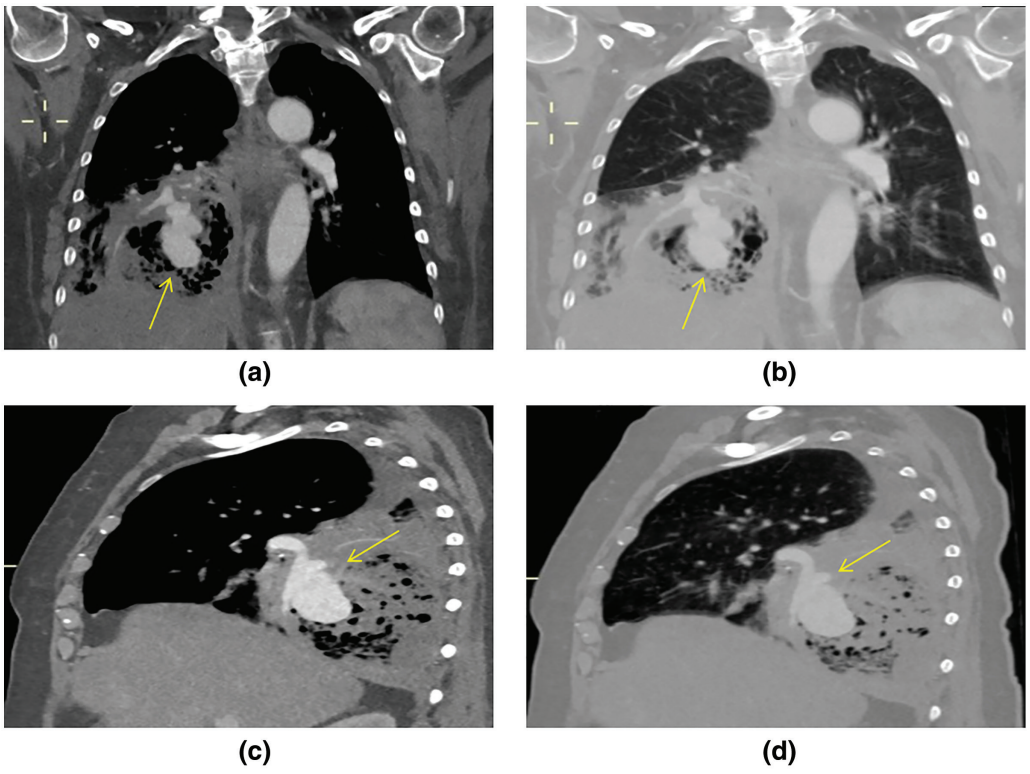


Figure 1 CT scan. CT scan showing in coronal (**a,b**) and sagittal (**c,d**) reconstructions showing the lower right pulmonary lobe almost completely replaced by colliquated metastases with evidence of a giant pseudoaneuryms (yellow arrows) of the posterior lower branch of the right pulmonary artery.

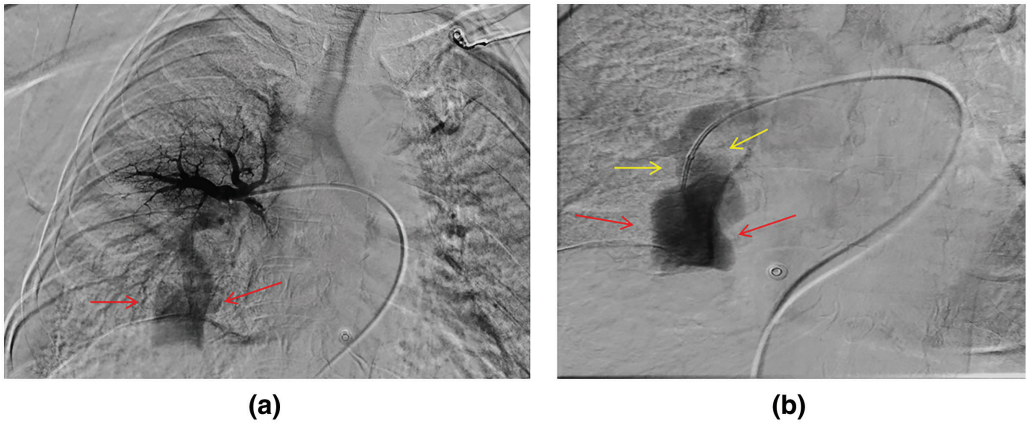


Figure 2 Digital Angiography. Selective angiography of the right pulmonary artery confirming the presence of a giant pseudoaneurysm (**a,b**) (red arrows) of its posterior lower branch; selective catheterisation of the pseudoaneurysm's sac (**b**) with evidence of the in-flow tract (yellow arrows).

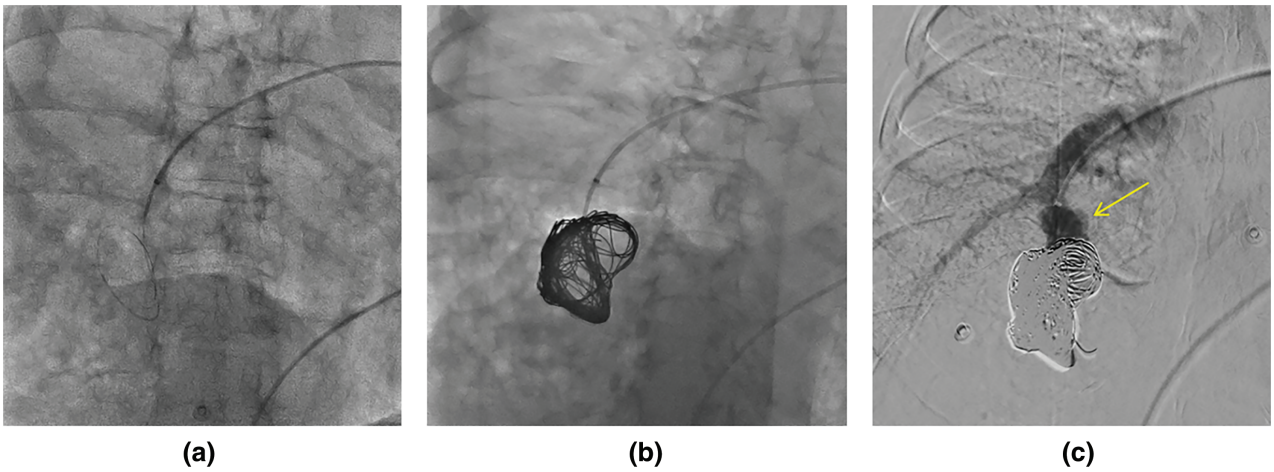


Figure 3 Digital Angiography. Selective catheterisation of the pseudoaneurysm's sac (**a,b**) with subsequent endovascular embolization, releasing multiple metallic microcoils; selective angiographic control (**c**) confirming successful endovascular exclusion of the pseudoaneurysm's sac with residual patency of its neck (**c**).

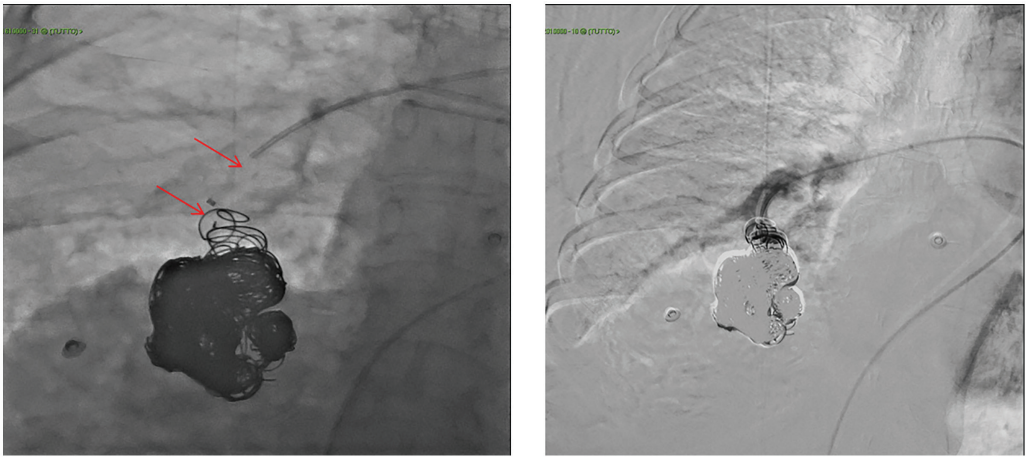
latter (Figure 4*a*) releasing an Amplatzer micro-vascular-plug (MVP-9Q, Medtronic, Inc. 710 Medtronic Parkway Minneapolis, MN 55432 USA).

Final angiographic control (Figure 4*b*) showed a complete exclusion of the pseudoaneurysm and its in-flow tract, with patency of the main trunk of right pulmonary artery and its remaining division branches. The patient was asymptomatic after the procedure, with significant improvement of serum hemoglobin values, and never experienced hemoptysis again; she was discharged 4 days after the embolisation.

DISCUSSION

Hemoptysis is the emission of red aerated blood from the mouth during a cough episode [5,10]. Massive haemoptysis is a blood loss of 300–600 mL over 24 hours and represents a distressing and life-threatening condition. Massive hemoptysis in 95% of cases originates from bronchial artery system, with pulmonary arteries accounting for 6%–11% of cases [5,7,10,11,12]. The most common etiology of hemoptysis from pulmonary artery system is represented by PAPs: it is a focal dilation of a pulmonary artery branch, involving only the adventice, with a higher risk of rupture than a true aneurysm. It constitutes a rare finding, often underdiagnosed by radiologists.

However, PAPs can be life-threatening if undiagnosed, due to a high mortality rate (50%) [5]. There are numerous potential etiologies of PAP: infection (tuberculosis, aspergillosis, necrotising pneumonia), iatrogenic (catheterisation of the pulmonary artery, lung radiofrequency ablation and surgical injury), trauma (penetrating or blunt injury), vasculitis (Bechet disease, Hughes–Stovin syndrome) and malignancy, usually primary lung cancer and rarely metastases [6–9,11,13–15].



(a) (b)

Figure 4 Digital Angiography. Endovascular embolisation **(a)** of the pseudoaneurysm's in-flow tract (neck) releasing metallic microcoils and its final sealing (red arrows) releasing an Amplatzer micro-vascular-plug; selective final angiographic control **(b)** confirming successful endovascular exclusion of the pseudoaneurysm's sac and its in-flow tract with patency of the main trunk of right pulmonary artery and its remaining division branches.

PAP due to primary lung cancer is rare and due to tumor necrosis: the pathological types of cancer involved in PAP are squamous-cell, small-cell and adenocarcinoma [6–9,15–17]. In the reported case, we described a rare giant PAP due to confluent colliquated mediastinal neuroendocrine carcinoma metastases, involving almost all the right lower pulmonary lobe. Patients affected by PAPs usually present with hemoptysis and hypoxemia, and sometimes experience chest pain too. Our patient, in fact, experienced in the last 2 days massive hemoptysis accompanied by severe dispnea. Chest radiography usually shows focal consolidation, solitary pulmonary nodules or multiple pulmonary nodules adjacent to the central or peripheral pulmonary vasculature [4,18]. Today, CT angiography represents the imaging modality of choice that not only establishes diagnosis, but, if performed with an appropriate timing of intravenous contrast, also helps to plan therapy with an endovascular approach. On CT angiography, PAPs appear as focal outpouchings of contrast medium adjacent to a pulmonary artery branch, showing the same contrast density as the pulmonary artery in all the dinamic phases of the study [1,2,4,11,19,20].

PAPs are more likely to rupture than true arterial aneurysm and, therefore, hemoptysis due to a ruptured PAP is often fatal and, in this way, it must be promptly recognised and treated [4]. Management of PAPs includes medical treatment, surgical approach and minimally invasive techniques. Antimicrobial therapy still keeps an important role in managing mycotic PAPs: empiric intravenous antimicrobial therapy targeting broad gram-negative and gram-positive coverage should be onset as soon as PAP is suspected. In addition, antifungal, antimycobacterial and antitreponemal therapy must be considered in immunocompromised patients [4].

An operative approach for PAPs is represented by open thoracotomy and aneurysm resection, with lobectomy for the involved pulmonary lobes. Surgical treatment, however, is associated with a high risk of morbidity and mortality, especially because patients affected by mycotic PAPs are usually acutely ill and with a poor pulmonary reserve [4,5,21,22,23]. Minimally invasive techniques, such as endovascular or percutaneous approaches, under CT scan, represent the elective initial therapy for these patients [5–9,15,16,24]. Urgent endovascular treatment is the preferred approach in managing hemoptysis due to PAPs, and such treatment should not be delayed: most PAPs can be successfully treated by endovascular procedures, mainly represented by endovascular embolisation [4,6–9,15,16,25,26]. These are the reasons why we decided to perform the endovascular embolisation in our patient and to attempt firstly a minimally invasive approach.

Many different embolic agents have been described, such as coils, Amplatzer vascular-plugs, detachable ballons, liquid agents (N-butyl cryanoacrylate, thrombin, Onyx), and stents [2,5–7,9,15,16,25]. The choice depends on the size of the PAP, the size of the neck, the location (proximal or distal) and the experience and preferences of the interventional radiologist [5,26]. Covered stent placement could be employed for endovascular treatment of central PAPs, especially if they are wide-necked, as the stent will maintain the permeability of the feeding artery. However, this may cause a risk of thrombosis or migration and, above all, eventual occlusion of other pulmonary artery branches, as seemed to be the case, after the preliminar angiographic

study, in our patient [4,5,6,17,18,24,27]. Moreover, potential graft infection, due to ongoing septic embolic phenomena, must be considered as a possible complication of covered stent placement and this approach should be avoided in patients with active bacteremia [4]. The target of the endovascular embolisation to pseudoaneurysms is both to fill the aneurysm's sac with embolic agents and to exclude the neck of the aneurysm from the circulation [28,29,30,31,32]. The embolisation of the proximal and distal neck is the most commonly reported endovascular approach because it is necessary to exclude both in-flow and out-flow tracts to avoid the risk of eventual late anterograde and retrograde reperfusion [28,33].

In the reported case, the giant pseudoaneurysm originated from a high-flow main division branch of the right pulmonary artery and had no out-flow vessels, but just a single in-flow tract, represented by its neck, which was, however, wide. The packing of the sac only is not enough as an endovascular approach, since it is necessary to seal its in-flow tract too, to avoid a later reperfusion of the sac due to the dislocation of the coils, already released, under the blood pressure of the main pulmonary artery [34]. Liquid agents in our patient were not indicated, as they could not have allowed a safe embolisation, due to the high-flow and wide-necked PAP, exposing the risk of a non-target embolisation [5,24,27]. For the reasons mentioned above, we decided to employ only mechanical agents, and not liquid agents, in our patient, and to perform first an endovascular embolization of the sac, to allow in this way a safer sealing of its wide in-flow tract by finally releasing the Amplatzer micro-vascular-plug.

CONCLUSIONS

PAPs are an uncommon, but potentially fatal, cause of hemoptysis; the most common etiology is represented by necrotising inflammatory processes, while less commonly they are due to to malignancy, more frequently primary lung cancer and rarely metastases. Since they are more likely to rupture than true arterial aneurysms, it is fundamental to promptly recognise and treat PAPs.

Endovascular embolisation represents a valid minimally invasive, safe and effective alternative treatment to the traditional surgical approach, with lower morbidity mortality and high success rates, expecially in those patients who are poor surgical candidates.

Ethics Statement

(1) All the authors mentioned in the manuscript have agreed to authorship, read and approved the manuscript, and given consent for submission and subsequent publication of the manuscript.
(2) The authors declare that they have read and abided by the JEVTM statement of ethical standards including rules of informed consent and ethical committee approval as stated in the article.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

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Author Contributions

All the authors substantially contributed to the study and manuscript writing.

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