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Surgical approach to a mycotic aneurysm of the pulmonary artery presenting with hemoptysis – A case report and a review of the literature

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ABSTRACT

INTRODUCTION: Mycotic aneurysms of the pulmonary arteries are very rare and have high mortality. Risk groups are intravenous drug users and patients with congenital heart disorders. The surgical approach varies due to a limited number of reported cases.

PRESENTATION OF CASE: We present a case of a mycotic aneurysm of the right pulmonary artery in a 56-year old man presenting with recurrent pneumonias, weight loss and hemoptysis.

DISCUSSION: There is often a diagnostic delay because of non-specific symptoms mimicking more common disorders. Treatment strategies include conservative management, surgery and endovascular treatment.

CONCLUSION: This report demonstrates a rare case of aneurysm of the pulmonary artery presenting with hemoptysis. For rapidly progressing proximal aneurysms of the pulmonary arteries, the midline surgical approach is recommended.

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1. Introduction

Aneurysms are irreversible vascular dilatations involving all three layers of the vessel wall [1]. Infections causing aneurysms, mycotic aneurysms, are estimated to represent only 1–3% of all arterial aneurysms and are rarely located in pulmonary arteries [2]. Patients may present with dyspnea, chest pain, cough, pulmonary hypertension, right ventricular failure and hemoptysis. Hemoptysis is estimated to be present in less than 10% of cases, however, when present the suspicion of a pulmonary aneurysm should be raised [1,2]. The non-specific presentation often causes diagnostic delay resulting in critical progression of the aneurysm.

We report here a case of successful surgical treatment for a mycotic aneurysm of the right pulmonary artery. The present case has been reported in line with the SCARE-criteria [3].

2. Presentation of case

A 56-year old man was admitted with recurrent pneumonias and symptoms of dyspnea and productive coughing for eight months, as well as hemoptysis and relapsing fever for the last three

months. Moreover, he had a weight loss of 12 kilos, excessive tiredness and persistently elevated infection parameters. A PET/CT scan showed pneumonic infiltrates in the upper and lower lobe of the right lung and an enlarged mass in the right hilum, interpreted as lymph nodes. Bronchoscopy and bronchoalveolar lavage revealed no signs of malignancy. The patient was treated with antibiotics but showed no signs of recovery during the following three months. An additional CT scan with contrast was performed suggesting the enlarged mass in the right hilum to be an aneurysmatic expansion of the right pulmonary artery (55 mm) indicating a mycotic aneurysm-formation with peripheral pulmonary embolisms to all three lobes of the right lung (Figs. 1 and 2). Following a multidisciplinary team meeting it was decided to manage the aneurysm conservatively with anticoagulants and antibiotics and repeat CT scan after 4 weeks. However, four days later an x-ray raised suspicion of progression of the aneurysm. An urgent CT scan confirmed progression of the aneurysm to 595 mm, as well as progression of pulmonary embolisms (Fig. 3). Transesophageal echocardiography (TEE) showed no signs of endocarditis or pulmonary hypertension. There was no confirmation of positive blood cultures. The treatment strategy was changed to a pneumonectomy of the right side.

A pneumonectomy of the right lung was performed through a median sternotomy. Intraoperatively, the tissue of the aneurysm was considered of good quality, which made it possible to resect the tissue in close proximity to the aneurysm. The operation was per-

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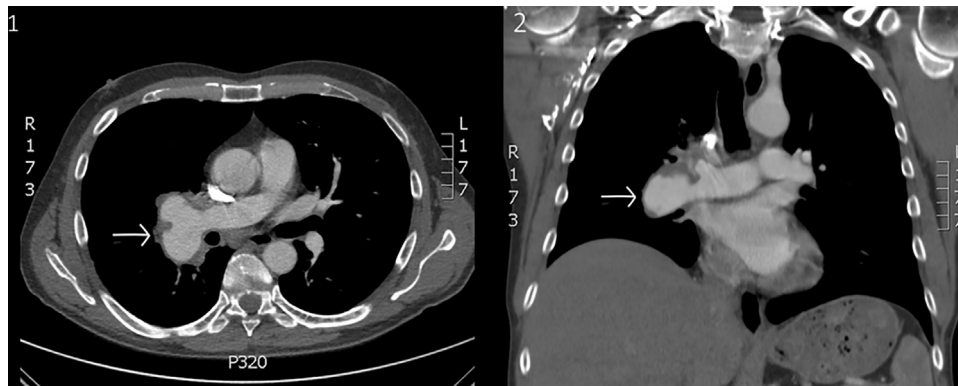


Fig. 1. 1) Cross-sectional CT with contrast. Arrow pointing at the mycotic aneurysm with thrombus-formation located on the right pulmonary artery; 2) Longitudinal CT contrast. Arrow pointing at the mycotic aneurysm.

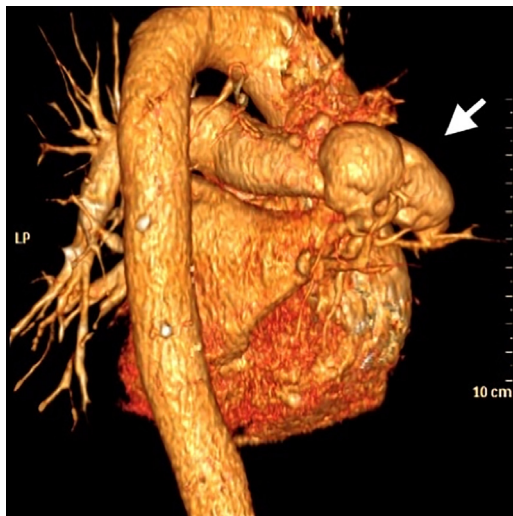


Fig. 2. CT reconstruction from a posterior-lateral view. Arrow pointing at the aneurysm on the right pulmonary artery.

formed without complications and the patient made an uneventful recovery and was discharged 15 days after surgery. Biopsies from the aneurysm revealed no microbiological focus, despite this the patient received Carbapenem for two weeks and Moxifloxacin for another two weeks postoperatively. Four months after the surgery, the patient revealed no symptoms or signs of infection and has started working again. Control CT scan shows no abscesses or new aneurysm formation.

3. Discussion

The case presented several diagnostic and management challenges. The enlarged mass in the right hilum found on X-ray was initially interpreted as enlarged lymph nodes. Due to malignancy-suspect symptoms such as weight loss, coughing and recurrent pneumonias, cancer was considered as a differential diagnosis, but was excluded on the subsequent PET/CT scan. Bronchoscopy and bronchoalveolar lavage revealed no signs of malignancy, and the patient was discharged with antibiotics. When the patient showed no signs of recovery after months, a CT scan was performed, and what was thought to be enlarged lymph nodes was now recognized as a pulmonary artery aneurysm with several septic thromboembolisms.

Relative few cases have been reported of aneurysms of the pulmonary arteries. Known risk factors are IV drug use, congenital heart disease such as ventricular septal defect (VSD), generalized

vasculitis, pulmonary hypertension, infection and immunosuppression [1]. Additional, according to Table 1 endocarditis seems to be a major predisposing factor for development of mycotic aneurysms. Mycotic aneurysms of the pulmonary arteries constitute a minority of the aneurysms at this site, and as seen in Table 1 they are commonly caused by suppurative bacterial infections such as staphylococci and streptococci or fungi including *Candida* and *Aspergilli* [4,5]. In the present case neither a bacteriological focus nor any of the risk factors mentioned above were present, however, since several septic thromboembolisms were located in the right lung, a mycotic aneurysm was a plausible diagnosis.

The proposed pathological mechanisms are i) direct spreading to a pulmonary artery from an adjacent focus of pulmonary infection, ii) ischemic injury to the pulmonary arterial wall as a result of infection of the vasa vasorum or iii) direct extension into a vessel wall from an intraluminal septic thromboembolus or from hematogenous spreading [1,4]. According to Table 1, the latter is the most frequent route of transmission with endovascular seeding originating from endocarditis, IV drug use, skin abscesses or pneumonia [1]. Since TEE showed no signs of endocarditis and all blood cultures were negative, in our case direct spread from a pulmonary focus or endovascular seeding from the pulmonary embolisms are the most likely pathological mechanisms.

Pulmonary angiography was previously gold standard, but recently MRI and CT with contrast have become the preferred diagnostic alternatives [4], which is confirmed in this case report. According to Bartter et al. the prognosis for mycotic aneurysms of the pulmonary arteries without interventional treatment is awful with mortality rates of 40–82% due to rupture [1]. Indeed, these mortality rates are confirmed in Table 1 showing a 63% (10 of 16 cases) mortality rate for patients not receiving surgical intervention, in contrast, patients receiving open surgical intervention with aneurysmectomy or removal of the affected part of the lung have a reduced mortality rate of 22% (2 of 9 cases). The management of these patients is difficult due to a lack of clear guidelines and sparse clinical experience. The proposed treatment strategies are diverse and include surgical interventions such as aneurysmectomy, lobectomy, pneumonectomy or embolotherapy [4,5]. Small mycotic aneurysms can also be treated conservatively, but regular CT scans are mandatory [6]. Less invasive approaches, e.g. lobectomy or segmental resection of lung parenchyma through a thoracotomy, are normally recommended, if feasible. However, for rapid progressing proximal aneurysms of the pulmonary artery, a midline approach through a sternotomy is to be recommended due to the possibility of central control in case of complications such as bleeding and the possibility to divide the structures more proximally in case of fragile tissue. Alternative nonsurgical interventional procedures, such as coil embolization or occlusion by detachable balloons might be

Table 1
Clinical and laboratory features of reported patients with pulmonary mycotic artery aneurysm.

Source, year	Sex	Age (y)	Predisposing factor	Systolic pulmonary arterial pressure (mmHg)	Hemoptysis	Infecting organisms	Surgical treatment	Survival
Pirani, 1949 [7]	M	10	Lymphoblastoma	NS	+	Negative	None	-
Lewisohn, 1957 [8]	M	64	Pulmonary endocarditis	NS	-	M. Tetragenus, S. Aureus, pneumococcus, proteus, Strep. Viridans	None	-
Goh, 1974 [9]	F	13	PDA, endocarditis	48	-	S. Pyogenes	Aneurysmectomy, closure of PDA	+
Goh, 1974 [9]	M	7	PDA, endocarditis	58	-	Negative	None	-
Gorodezky, 1974 [10]	F	18	PDA, endocarditis	NS	+	Streptococcus	None	-
Singer-Jordan, 1980 [11]	M	23	Ventriculoatrial shunt	NS	+	Propionibacterium acnes	Pneumonectomy	+
Gelfand, 1981 [12]	M	23	Drug abuse, ventriculoatrial shunt due to hydrocephalus	NS	+	Propioni organisms, S. Epidermidis, Strep. Viridans	Pneumonectomy	+
Choyke, 1982 [13]	F	12	Corrected tetralogy of Fallot, tricuspid endocarditis	NS	-	CNS, Aspergillus	Tricuspid valve replacement	-
SanDretto [14]	F	36	Drug abuse, endocarditis	NS	-	CNS	None	+
Navarro, 1984 [15]	F	34	Drug abuse	NS	-	NS	None	-
Navarro, 1984 [15]	M	33	Drug abuse, tricuspid endocarditis	NS	-	S. Aureus, Streptococcus, Kkebsiella, Diphtheroids	None	-
Navarro, 1984 [15]	F	27	Drug abuse	NS	-	S. Aureus	None	-
Caplin, 1985 [16]	F	20	Closure of PDA	NS	-	S. Aureus	Closure of PDA, ligation of aneurysm	+
Morgan, 1986 [17]	M	20	Drug abuse	NS	+	S. Aureus	None	-
Roush, 1988 [18]	M	62	Tricuspid endocarditis	NS	-	S. Aureus, C. Albicans	Tricuspid valve replacement	-
Vargas-Barron, 1992 [19]	M	11	PDA, coarctation, vegetations on pulmonary artery	NS	-	S. Aureus	Correction of coarctation, closure of PDA, aneurysmectomy	+
Chung, 1995 [20]	F	58	None	NS	-	None	None	+
Benveniste, 1998 [21]	F	28	HIV, tricuspid and pulmonary endocarditis, drug abuse	60	+	S. Aureus and Strep. Oralis	Lobectomy	-
McLean, 1998 [6]	M	28	Drug abuse	NS	-	S. Aureus	None	+
Lawrenson, 1999 [22]	F	2	Tetralogy of Fallot, Blalock Taussig shunt, pulmonary endocarditis	NS	-	Strep. Viridans	Pulmonary valve replacement	+
Lertsapcharoen, 2002 [23]	F	9	PDA, endocarditis	NS	+	Streptococcus	Closure of PDA	+
Dransfield, 2003 [24]	F	49	None	NS	-	Str. viridans	None	-
Bozkurt, 2003 [25]	F	6	VSD, tricuspid endocarditis	NS	-	S. aureus	Lobectomy, plication of aneurysm, closure of VSD, removal of vegetations of tricuspid valve	+
Kim, 2004 [5]	M	71	None	NS	+	Actinomyces	Coil embolization	-
Greillier, 2005 [26]	M	53	Acute myeloid leukemia	NS	-	Aspergillus Fumigatus	None	-
Saida, 2007 [27]	F	49	Infection of pacemaker	NS	-	S. Aureus	None	+
Takao, 2007 [28]	F	21	VSD, endocarditis	NS	-	NS	Coil embolization	+
Wilson, 2008 [29]	F	22	Drug abuse, pulmonary endocarditis	NS	-	S. Aureus	None	+
Sever, 2010 [30]	M	49	Hairy cell leukemia, dental abscess	NS	+	Aspergillus	Lobectomy	-
Al Banna, 2011 [4]	M	48	Pericardial tamponade	27	-	C. Albicans	None	+
Groner, 2012 [31]	M	4	DiGeorge syndrome with operated tetralogy of Fallot, xenograft endocarditis	NS	-	C. Hominis	Coil embolization	-
Papaioannou, 2014 [2]	F	23	Drug abuse	NS	+	S. Aureus	Lobectomy	+
Toganel, 2014 [32]	M	2 months	Previous CVK, endocarditis	NS	+	C. Lusitaniae	Removal of thrombus in right ventricular outflow tract	+
Luo, 2015 [33]	F	29	PDA, pulmonary endocarditis	NS	-	NS	Closure of PDA	+
Calais, 2017 [34]	M	36	Endocarditis after Ross procedure	NS	-	NS	Pulmonary valve replacement	+
Knowles, 2017 [35]	F	16	Operated Tetralogy of Fallot, endocarditis	NS	-	S. Aureus	Surgical revision of conduit	+
Piracha, 2018 [36]	M	68	Chronic right upper lobe cavity	NS	+	Negative	Coil embolization	-
Srinivasa, 2018 [37]	F	13	DiGeorge syndrome, repair of truncus arteriosus, pulmonary artery hypoplasia, endocarditis	NS	-	Staphylococcus	Balloon occlusion	-

PDA: patent ductus arteriosus, VSD: ventricular septal defect, CVK: central venous catheter, NS: not specified, CNS: coagulase-negative staphylococcus. A search of the PubMed database was undertaken on 05/07/18 with the search terms (“Aneurysm, Infected” [Mesh] AND “Pulmonary Artery” [Mesh]). 89 articles were returned. Articles in non-english, articles regarding pseudoaneurysms and non-relevant articles were excluded, which left 40 articles. Of these, 13 were unable to be accessed. The remaining 27 articles were included in our paper. Furthermore, 8 articles were found via references, which resulted in a total of 35 papers included in the review.

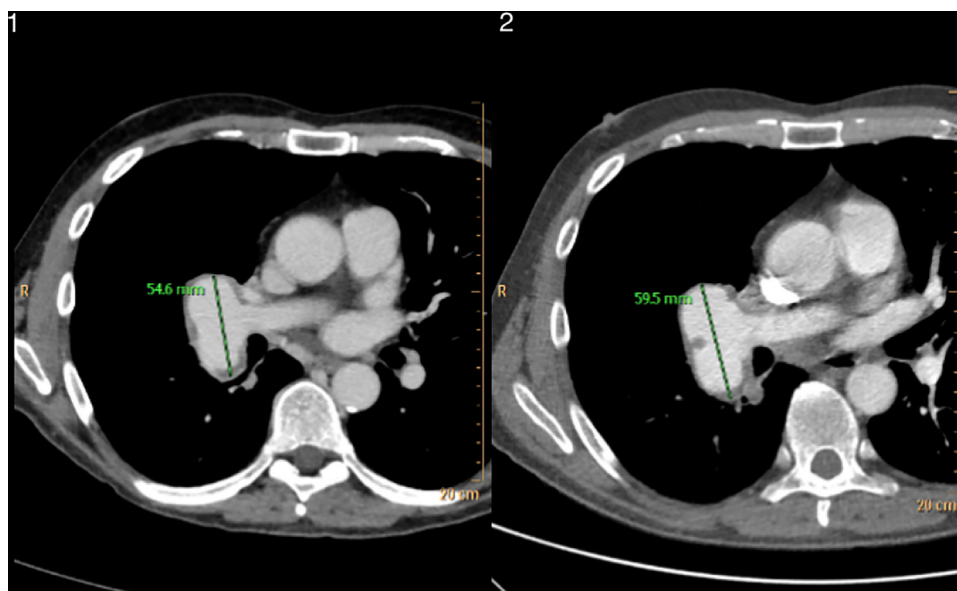


Fig. 3. 1) CT with contrast showing a mycotic aneurysm of the right pulmonary artery measuring 54.6 mm; 2) CT with contrast showing progression of the mycotic aneurysm measuring 59.5 mm.

used for peripheral pulmonary aneurysms, multiple bilateral pulmonary aneurysms or in patients who may not tolerate surgery [6]. In our case, the midline approach was chosen due to the central location and the rapid progression of the aneurysm, which indicated an unstable vessel wall with increased risk of hemorrhage. According to the literature the presence of hemoptysis appears to be a significant marker of instability of the lesion and a strong indicator of prompt intervention [1].

4. Conclusion

The diagnosis should be suspected when patients present with hemoptysis or signs of pulmonary hypertension, but also in the presence of more vague symptoms, especially in specific patient groups. The presence of hemoptysis and progression of the mycotic aneurysm should alert the clinician to consider a more aggressive treatment like aneurysmectomy or removal of the affected part of the lung. A midline approach is recommended in case of rapidly developing proximal aneurysms of the pulmonary artery to avoid potential complication of catastrophic bleeding and enable better surgical control.

Conflict of interest

No conflict of interest declared.

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Ethical approval

Consent from the patient is sufficient and ethical approval is not needed in Denmark.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy

of the written consent is available for review by the Editor-in-Chief of this journal on request.

Author contribution

All authors dr. Terp, dr. Højsgaard, dr. de Paoli and dr. Benhassen have taken part in conception of the study, drafting and revising the whole manuscript critically. All authors have given their final approval of the manuscript upon submission.

Registration of research studies

None.

Guarantor

Leila Louise Benhassen.

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