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CASE REPORT

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Pulmonary artery pseudoaneurysm after thoracic radiation therapy: A case report and review of the literature

Gerdinique (G. C.) Maessen[1] | Thijs (T. W.) Hoffman[1] | Lidwien (L.) Graat-Verboom[2] | Marc (M.) van Leersum[3] | Hans-Jurgen (J. J.) Mager[1]

[1]Pulmonology, St. Antonius Ziekenhuis, Nieuwegein, The Netherlands

[2]Pulmonology, Maxima Medisch Centrum, Eindhoven, The Netherlands

[3]Radiology, St. Antonius Ziekenhuis, Nieuwegein, The Netherlands

Correspondence
Gerdinique (G. C.) Maessen, Pulmonology, St. Antonius Ziekenhuis, Koekoekslaan 1, 3435 CM Nieuwegein, The Netherlands.
Email: nikimaessen@kpnmail.nl

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Abstract

Pulmonary artery pseudoaneurysm (PAP) is a rare cause of hemoptysis. Potential causes include trauma, infection, or medical interventions. There is a risk of rupture, which is associated with a high mortality rate. We describe a 72-year-old patient, with a past medical history of a lung carcinoma for which she was treated with chemoradiotherapy 6 years prior, who presented with hemoptysis. She was hemodynamically stable and there were no other complaints. CT angiography of the thorax showed a PAP originating from a branch of the right pulmonary artery in the previously irradiated area. The patient was successfully treated by an embolization with plugs. Treatment of lung carcinoma with chemoradiotherapy can result in the development of a PAP. Clinicians should be aware of this complication, even years after the therapy. In literature, only a few cases of PAP in patients treated with (chemo)radiotherapy for lung cancer are described, with a maximum interval up to 7 years.

KEYWORDS

case report, lung carcinoma, pulmonary artery pseudoaneurysm, radiotherapy

INTRODUCTION

Haemoptysis may be the presenting symptom of rare but potentially life-threatening pulmonary vascular abnormalities such as a pulmonary artery pseudoaneurysm (PAP). We present the case of a patient who was still followed up after chemoradiotherapy treatment for lung cancer.

CASE REPORT

A 72-year-old woman presented at the emergency department with haemoptysis, which had occurred acutely during a cough. She denied having chest pain, dyspnoea, fever, or malaise, and she did not report any other complaints recently. Her past medical history included chronic obstructive pulmonary disease (Gold category 3), a transient ischemic attack, and lung cancer in the right upper lobe (squamous cell carcinoma, TNM stage T4N1M0) which had been successfully treated with sequential chemoradiotherapy 6 years ago. Her current medication included inhalers (beclometasone dipropionate/formoterol fumarate dihydrate/glycopyrronium), maintenance antibiotic therapy (azitrhomycin), and clopidogrel.

On physical examination, the patient was hemodynamically stable, and did not appear dyspnoeic. She coughed up small amounts of bright red blood. Her oxygen saturation was 95% on room air, and auscultation of the lungs revealed diffuse crackles over both lungs. Laboratory investigations revealed a haemoglobin of 8.3 mmol/L (normal range: 7.0–9.8), a CRP of 82 mg/L (normal: <10), and leukocytes of 9.7 $\times$ 10^9/L (normal range: 3.7–10.9). A chest radiograph demonstrated a pre-existing volume loss of the right hemithorax with a small amount of pulmonary oedema and minimal infiltrations in the right lower lobe.

The patient was admitted to the inpatient ward with a working diagnosis of pneumonia. The differential diagnosis included vascular abnormalities or recurrent lung carcinoma. Clopidogrel was stopped and therapy with tranexamic acid and amoxicillin/clavulanic acid was initiated. A bronchoscopy was performed and showed an active bleeding

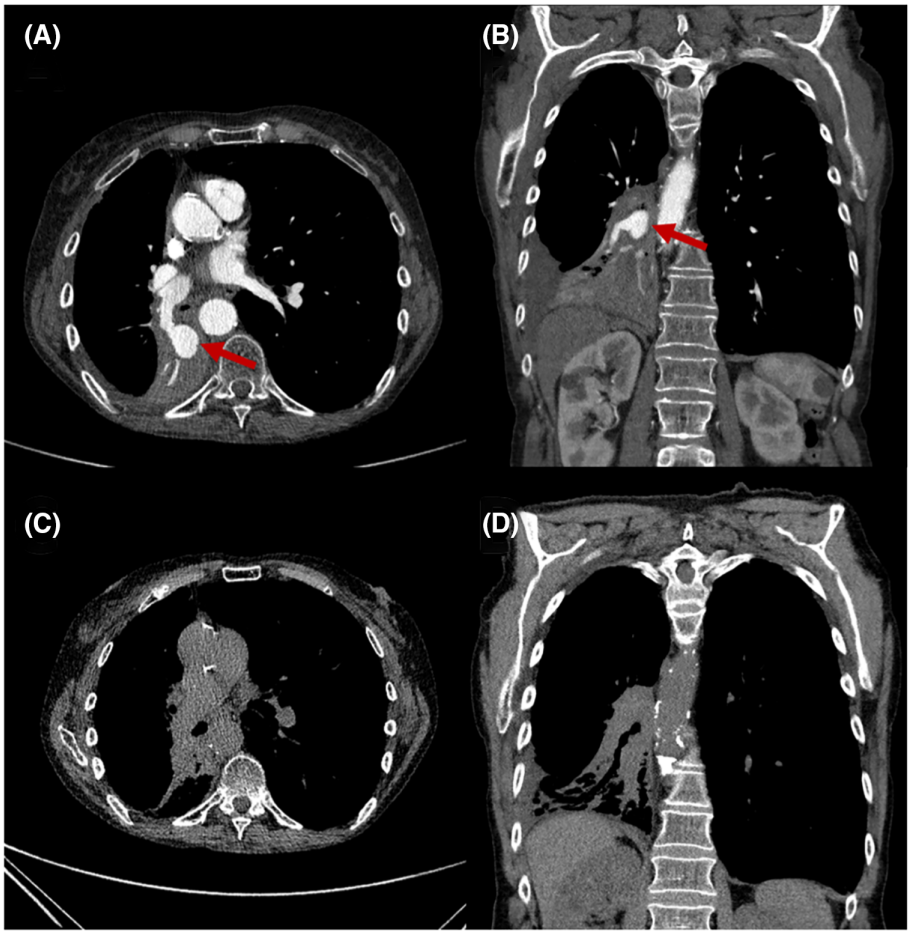

FIGURE 1 (A) CT of the chest with intravenous contrast in the pulmonary circulation, performed when the patient presented with haemoptysis, axial plane. This reveals a pseudoaneurysm originating from the right lower lobe branch of the pulmonary artery. (B) CT of the chest with intravenous contrast in the pulmonary circulation, performed when the patient presented with haemoptysis, coronal plane. This reveals a pseudoaneurysm originating from the right lower lobe branch of the pulmonary artery. (C) CT scan of the chest without intravenous contrast, 4 months prior to the current episode, axial plane, on which the pseudoaneurysm cannot be clearly seen. (D) CT scan of the chest without intravenous contrast, 4 months prior to the current episode, coronal plane, on which the pseudoaneurysm cannot be clearly seen.

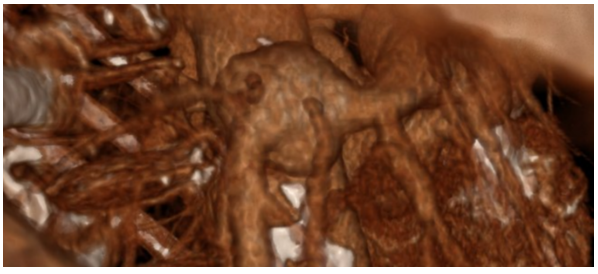

FIGURE 2 Three-dimensional image of the pseudoaneurysm.

in the right lower lobe, which stopped after suction and rinsing with xylometazoline. Computed tomography angiography (CTA) of the chest showed a 2.7 × 2 × 2.4 cm pseudoaneurysm originating from a branch of the right pulmonary artery (Figures 1A,B and 2). In addition, the CTA revealed slender bronchial arteries, obvious emphysema, volume loss of the right hemithorax caused by chemoradiotherapy, and atelectasis and obliteration of the right lower lobe, which were thought to be secondary to blood clots.

The patient was transferred to another hospital for further treatment. A successful embolization of the PAP with three IMPEDE plugs was performed (Figure 3). Afterwards, the patient had no more complaints of haemoptysis. The tranexamic acid was ceased and the clopidogrel resumed. The patient could be discharged home and has not had recurrent haemoptysis in the 3 months after this episode.

DISCUSSION

Patients with moderate to massive haemoptysis mostly suffer from bleeding of the systemic circulation (bronchial artery). In 5%–10% however, the source of bleeding is the pulmonary circulation, potentially caused by a PAP.[1] A PAP is a focal dilatation of the pulmonary artery, not involving all three but only two arterial wall layers, in contrast to a true aneurysm.[2,3] Histologically, a PAP is confirmed by the

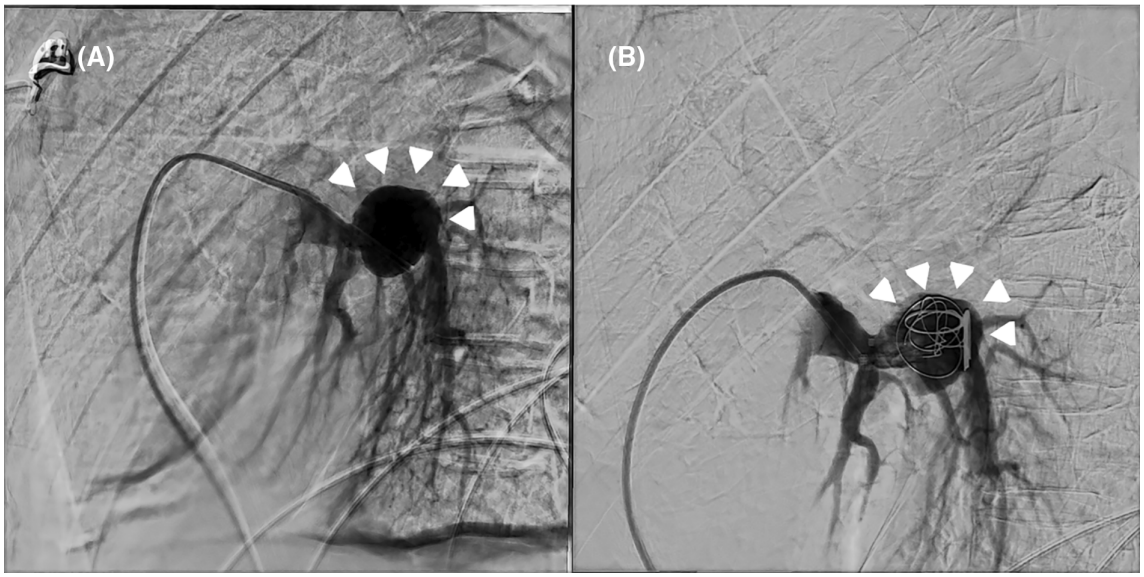

FIGURE 3 (A) Pulmonary artery angiography, where the right lower lobe branch of the pulmonary artery is selectively visualized, revealing a pulmonary artery pseudoaneurysm. (B) Pulmonary artery angiography, where the right lower lobe branch of the pulmonary artery is selectively visualized, revealing embolization plugs in the pulmonary artery pseudoaneurysm.

TABLE 1 Causes of acquired pulmonary artery pseudoaneurysms.

Causes of acquired pulmonary artery pseudoaneurysms	
Iatrogenic	Placement of pulmonary catheter (Swan-Ganz)
	Right heart catheterization
	Pulmonary angiograph
	Intubation
	Lobectomy
	Radiation
	Radiofrequency ablation
Traumatic	Stab wound, gunshot, or non-penetrating trauma
Infectious	Tuberculosis (Rasmussen aneurysm)
	Syphilis
	Mycotic (secondary to bacterial endocarditis)
Neoplasm	Primary lung carcinoma (usually squamous cell carcinoma)
	Metastasis (including sarcoma, breast carcinoma)
Vasculitis	Behçet's disease
	Takayasu arteritis
Other	Chronic thromboembolic pulmonary hypertension
	Bronchiectasis
	Pulmonary fibrosis

absence of a three-layer structure of the vessel wall. However, most PAPs are diagnosed based on radiological appearance and clinical context, as is the case with our patient. The clinical presentation may vary from an asymptomatic PAP that is discovered coincidentally on imaging studies to massive haemoptysis.[3]

CTA with contrast in the pulmonary circulation is the gold standard for diagnosis, follow-up, and planning of a potential intervention of a PAP. Doctors should always be aware of a PAP as potential cause of haemoptysis. An American study found that in half of the cases a PAP was missed at the first evaluation by a radiologist, even when haemoptysis was mentioned in the request for the CTA.[4] Our patient had been followed up since she received chemoradiotherapy as treatment for lung cancer 6 years ago, however imaging during this follow-up consisted of CT scans without intravenous contrast, which also retrospectively did not show the PAP.

Most PAPs in Western countries are secondary to trauma or medical interventions.[5] Other possible causes are infection, vasculitis, and neoplasm (Table 1).[3] The prognosis depends on the cause of the PAP, but mortality can be more than 50% in case of a rupture.[6] For patients with underlying lung cancer, survival rates of 46%–67% during 1–3 months after embolotherapy of the PAP are reported in the literature.[7]

In case of an asymptomatic, small PAP, a wait-and-see policy is justified since spontaneous regression can occur.[8] Haemoptysis is an indication for treatment. Supportive therapy could be given in the form of antifibrinolytics. For definitive treatment, endovascular therapy is preferred over surgery, though the latter may be useful in some cases.[2,5,7] Embolization is the most successful and described intervention, with a success rate of 75%.[4,9] In contrast to endovascular treatment of haemoptysis originating from the bronchial artery, embolization of the pulmonary artery is performed with plugs or coils, and not with microparticles. Even so, in case of a PAP, additional embolization of branches originating from the bronchial artery may be necessary.[10]

Our patient had no history of trauma or infection, and there were no other clues to any aetiology other than the chemoradiotherapy that she received in the past. Her PAP originated from branches in the irradiated area, and likely

TABLE 2 Cases with pulmonary artery pseudoaneurysm after (chemo)radiotherapy treatment for lung carcinoma.

Author, year and reference	Age	Sex	Diagnosis and treatment	Interval between (start of) treatment and PAP discovery	PAP size	Treatment for PAP	Outcome
Millet et al.[15] 2008	59	M	Recurrence of squamous cell carcinoma (initially treated with lobectomy), CRT	1 week	Not described	Embolization	Death (2 days later due to septic shock)
Kim et al.[16] 2015	64	M	Squamous cell carcinoma, CRT	4 months	48 × 32 mm	Conservative care	Death
Chen et al.[4] 2017	68	M	Squamous cell carcinoma, RT	5–9 months	Not described	Embolization with coils	Not described
Sabir et al.[17] 2017	70	F	Suspicion of malignancy (20 years after adenocarcinoma treated with lobectomy), RT	2 years	Not described	Embolization with coils and stent graft	Recurrence of haemoptysis 2 days later, start palliative care
Petsatodis et al.[18] 2019	64	F	SCLC, CRT	6 months	Not described	Placement transluminar stent graft	Survival
Fukuda et al.[11] 2020	65	M	Squamous cell carcinoma, RT	1 month	Not described	No intervention (death before scheduled embolization)	Death
Kang et al.[19] 2020	64	M	Squamous cell carcinoma, RT	6 days	65 × 37 mm	Embolization with 3 tungsten coils	Survival (5-month follow-up)
Doyle et al.[20] 2021	43	M	Pulmonary sarcoma, lobectomy, (neo)adjuvant CRT and RFA	7 years	Not described	Placement aortic stent graft system	Survival
Lutz et al.[21] 2021	75	M	Adenocarcinoma, lobectomy and (neo)adjuvant CRT	7 years	41 × 22 × 33 mm	Embolization with 8 coils *Second attempt 4 days later*: Onyx within coil packing	Persistent haemoptysis *After second embolization*: survival
Sommer et al.[22] 2022	58	M	NSCLC (no subtype), CRT	'Recently'	Not described	Embolization with various types of coils	Survival
Present case	72	F	Squamous cell carcinoma, CRT	6 years	27 × 20 × 24 mm	Embolization with three IMPEDE plugs	Survival

Abbreviations: CRT, chemoradiotherapy; (N)SCLC, (non-)small cell lung carcinoma; RFA, radiofrequency ablation; RT, radiation therapy.

represents a radiotherapy-induced PAP. However, recurrence of the lung cancer cannot be definitively ruled out. An earlier case report described a fatal case of bleeding from a post-radiation PAP in whom obduction showed invasion of both inflammatory and malignant cells.[11] Although several studies on PAP have been reported, only 10 cases concerned patients treated with (chemo)radiotherapy for lung cancer. A summary of these cases is presented in Table 2. Most patients were middle-aged and older males, and all patients presented with haemoptysis like our patient. The PAP was mostly discovered several months after the patient started or received (chemo)radiotherapy, but in some cases the interval was more than 5 years. As mentioned before, embolization is the most used intervention, however these reports show that placement of a stent graft is also performed occasionally. It seems that embolization occurs mostly with coils; our patient is the first of this group reported to have been embolized with plugs. Embolization with vascular plugs is described previously in other cases of iatrogenic PAPs.[12] A recent review of embolization of PAPs stated that the choice for embolizing material depends largely on the experience of the interventional radiologist.[13] An advantage of plugs is the mechanical obstruction of the feeding artery, a disadvantage is the need for a relatively large catheter to place the plug, which may be difficult in the case of a tortuous feeding artery. There are no studies comparing outcomes of patients with a PAP treated with either coils or plugs. For hemoptysis related to lung tumours in general however, a recent study showed no difference in recurrence rate of survival between these groups.[14] Since not all studies included in this review provide information on the size of the PAP and follow-up, it is not possible to compare the therapies and outcomes for post-radiation PAP as well.

In conclusion, a PAP is a potential cause of moderate to massive haemoptysis. Trauma, infection, or cancer can result in the development of a PAP. Also, iatrogenic causes, such as previous (chemo)radiotherapy have to be considered. It is important to be aware of this complication in

patients with a history of (chemo)radiotherapy. The gold standard for diagnosis of a PAP is a CTA of the chest with contrast in pulmonary circulation. Endovascular embolization is an effective treatment in most cases.

AUTHOR CONTRIBUTIONS

GCM: Conception of the work, analysis and interpretation of data; drafting, updating and literature search, and final approval of the version to be published. **TWH**: Conception of the work, acquisition, analysis and interpretation of data, drafting and reviewing it for important intellectual content, and final approval of the version to be published. **LGV**: Acquisition of data, reviewing it for intellectual content, and final approval of the version to be published. **MvL**: Acquisition of data, reviewing it for intellectual content, and final approval of the version to be published. **JJM**: Interpretation of data, reviewing it for intellectual content, and final approval of the version to be published.

CONFLICT OF INTEREST STATEMENT

None declared.

DATA AVAILABILITY STATEMENT

The authors confirm that the data supporting the findings of this study are available within the article.

ETHICS STATEMENT

The authors declare that appropriate written informed consent was obtained for the publication of this manuscript and accompanying images.

ORCID

Gerdinique (G. C.) Maessen https://orcid.org/0009-0007-2387-3305

REFERENCES

1. Khalil A, Fedida B, Parrot A, Haddad S, Fartoukh M, Carette MF. Severe hemoptysis: from diagnosis to embolization. Diagn Interv Imaging. 2015;96(7–8):775–88. https://doi.org/10.1016/j.diii.2015.06.007
2. Bhatty D, Srivali N. Pulmonary artery pseudoaneurysm: a rare cause of hemoptysis. QJM. 2020;113(5):351–2. https://doi.org/10.1093/QJMED/HCZ316
3. Guillaume B, Vendrell A, Stefanovic X, Thony F, Ferretti GR. Acquired pulmonary artery pseudoaneurysms: a pictorial review. Br J Radiol. 2017;90(1073):20160783. https://doi.org/10.1259/BJR.2016078
4. Chen Y, Gilman MD, Humphrey KL, Salazar GM, Sharma A, Muniappan A, et al. Pulmonary artery Pseudoaneurysms: clinical features and CT findings. AJR Am J Roentgenol. 2017;208(1):84–91. https://doi.org/10.2214/AJR.16.16312
5. Lafita V, Borge MA, Demos TC. Pulmonary artery pseudoaneurysm: etiology, presentation, diagnosis, and treatment. Semin Intervent Radiol. 2007;24(1):119–23. https://doi.org/10.1055/S-2007-971202
6. Ungaro R, Saab S, Almond CH, Kumar S. Solitary peripheral pulmonary artery aneurysms. Pathogenesis and surgical treatment. J Thorac Cardiovasc Surg. 1976;71(4):566–71. https://doi.org/10.1016/s0022-5223(19)40180-3
7. Tantrige P, Johnston E, Garzillo G, Huang DY. Pulmonary artery pseudoaneurysm embolisation to treat massive haemoptysis due to metastatic oropharyngeal squamous cell carcinoma. BMJ Case Rep. 2019;12(6):e230283. https://doi.org/10.1136/BCR-2019-230283
8. You CK, Whatley GS. Swan-Ganz catheter-induced pulmonary artery pseudoaneurysm a case of complete resolution without intervention. Can J Surg. 1994;37:420–4.
9. Bussières JS. Iatrogenic pulmonary artery rupture. Curr Opin Anaesthesiol. 2007;20(1):48–52. https://doi.org/10.1097/ACO.0B013E32801158A9
10. Shin S, Shin TB, Choi H, Choi JS, Kim YH, Kim CW, et al. Peripheral pulmonary arterial pseudoaneurysms: therapeutic implications of endovascular treatment and angiographic classifications. Radiology. 2010;256(2):656–64. https://doi.org/10.1148/RADIOL.10091416
11. Fukuda Y, Homma T, Uno T, Murata Y, Suzuki S, Shiozawa E, et al. Fatal rupture of pulmonary artery pseudoaneurysm after thoracic radiation therapy against lung squamous cell carcinoma: a case report and literature review. Clin Case Rep. 2020;9(2):737–41. https://doi.org/10.1002/CCR3.3647
12. Burrel M, Real MI, Barrufet M, Arguis P, Sánchez M, Berrocal L, et al. Pulmonary artery pseudoaneurysm after Swan-Ganz catheter placement: embolization with vascular plugs. J Vasc Interv Radiol. 2010;21(4):577–81. https://doi.org/10.1016/j.jvir.2009.12.399
13. Fontana F, Piacentino F, Curti M, Ierardi AM, Coppola A, Macchi E, et al. Pulmonary artery pseudoaneurysms embolization: bicentric experience and review of the literature. J Clin Med. 2023;12(11):3796. https://doi.org/10.3390/jcm12113796
14. Claudinot A, Douane F, Morla O, Perret C, Neveu M, Thouveny F, et al. Pulmonary artery embolization in the management of hemoptysis related to lung tumors. J Pers Med. 2023;13(11):1597. https://doi.org/10.3390/jpm13111597
15. Millet A, Morel H, Heautot JF, Corre R, Curran Y. Faux anévrisme de l'artère pulmonaire sur récidive de cancer [false aneurysm of the pulmonary artery secondary to a cancer recurrence]. Presse Med. 2008;37(12):1817–9. French. https://doi.org/10.1016/j.lpm.2007.10.021
16. Kim JH, Han SH. A pulmonary artery pseudoaneurysm caused by concurrent chemoradiation therapy for lung cancer. Pak J Med Sci. 2015;31(1):220–2. https://doi.org/10.12669/pjms.311.6001
17. Sabir R, Umar M, Khan A, Gurung P. Pulmonary artery pseudoaneurysm following stereotactic body radiation therapy. Am J Respir Crit Care Med. 2017;195:A6642. https://doi.org/10.1164/ajrccmconference.2017.C80I
18. Petsatodis E, Giankoulof C, Glanais S. Endovascular treatment of a pulmonary artery pseudoaneurysm with a stent graft. CIRSE 2019 abstracts. Cardiovasc Intervent Radiol. 2019;42(3):171. https://doi.org/10.1007/s00270-019-02282-x
19. Kang T, Kang MJ. Pulmonary artery pseudoaneurysm showing rapid growth in a patient with lung cancer. Radiol Case Rep. 2020;15(11):2144–8. https://doi.org/10.1016/j.radcr.2020.07.077
20. Doyle J, Mhandu P. Late presentation of pulmonary artery pseudoaneurysm following bronchovascular sleeve resection of lung. BMJ Case Rep. 2021;14(7):e243294. https://doi.org/10.1136/bcr-2021-243294
21. Lutz N, Grange S, Sanson C, Grange R. Successful embolization of giant pulmonary artery pseudoaneurysm using coils and ethylene vinyl alcohol copolymer (onyx). Radiol Case Rep. 2021;16(5):1068–71. https://doi.org/10.1016/j.radcr.2021.02.029
22. Sommer CJ, O'Neal D, Andrews RH. Embolic pneumonectomy for the treatment of proximal pulmonary artery Pseudoaneurysm. Cureus. 2022;14(7):e26487. https://doi.org/10.7759/cureus.26487

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