Article published online: 2020-12-22

Letter to the Editor

Pulmonary artery pseudoaneurysm secondary to metastatic breast cancer

DOI: 10.4103/sajc.sajc_157_17

Dear Editor,

An aneurysm is defined as a focal dilatation of a blood vessel involving all three layers of the vessel wall: tunica intima, media, and adventitia.[1] In contrast, a pseudoaneurysm is caused by disruption of the vessel wall, causing blood to leak through and only contained by adventitia or perivascular soft tissue.[1]

Pulmonary artery pseudoaneurysm (PAP) arising from neoplasm is rare. In previous literature, one case of epithelioid hemangioendothelioma, five cases of sarcoma relating to hemangiopericytoma, angiosarcoma and leiomyosarcoma, as well as 12 cases of primary lung cancer have been identified.[2-16] Herein, we describe the first published case of a PAP originating from metastatic breast cancer.

A 62-year-old female ex-smoker presented to the emergency department after a 2-week history of increasing dyspnea and dry cough. She has a history of breast cancer, histologically proven to be triple negative, invasive ductal carcinoma with sarcomatoid features and previously treated with mastectomy and adjuvant chemotherapy. Previously computed tomography (CT) scans have revealed skeletal, nodal, and pulmonary metastases, including T8 vertebral metastasis requiring radiotherapy, surgical debulking, and pedicle screw fixation vertebroplasty due to spinal cord compression. She denied any hemoptysis, chest pain, or fever. Her last cycle of chemotherapy was 2 months prior to presentation, ceased before completion due to poor response. However, she was on dexamethasone for the management of her skeletal metastases.

On examination, she was afebrile but tachycardic (heart rate up to 120 beats/min), mildly tachypneic (respiratory rate of 20 breaths/min), hypertensive (blood pressure 180/100 mmHg), and severely hypoxic (oxygen saturation of 66% on room air). Apart from decreased breath sounds in the left hemithorax, physical examination was unremarkable. Chest radiograph showed an interval growth of known left-sided mediastinal mass as well as interval development of moderate volume bilateral pleural effusions.

CT pulmonary angiogram (CTPA) was subsequently performed to exclude pulmonary emboli. No pulmonary emboli were detected. However, disease progression was evident with increased number and the size of pulmonary metastatic lesions as well as bilateral moderate volume pleural effusions. A previously imaged right middle lobe metastasis [Figure 1] had increased in size measuring up to 2.7 cm (previously 1 cm) and now exhibited central arterial contrast enhancement, with a stem arising directly from the medial segmental branch of the right middle lobe pulmonary artery [Figure 2]. Pseudoaneurysm size measured up to 1.3 cm in diameter. Findings are compatible with a malignant PAP.

In light of the patient's widespread, treatment-resistant metastatic disease, the decision to initiate palliative care was made with no further interventional options deemed suitable for the pseudoaneurysm. She was treated with pleurocentesis for symptomatic relief and subsequently discharged home.

PAP is a rare condition which can be congenital or acquired.[17] The leading cause of PAP is iatrogenic, such as following Swan-Ganz catheter insertion, cardiac catheterization, chest tube insertion, conventional angiography, radiofrequency ablation, biopsy, and surgical resection.[1,17-19] Trauma is the second most common cause.[1,6,13] Other causes include vasculitis (Behcet's disease and Takayasu arteritis), infection (tuberculosis, pyogenic infection, mucormycosis, subacute bacterial endocarditis, aspergillosis, necrotizing pneumonia, and candidiasis), and neoplasm.[17,20]

(Continue on page 26...)

(Letter to the editor continue from page 20...)

To the best of our knowledge, PAP is rarely associated with malignancy. Within the current literature, we have identified only one case of epithelioid hemangioendothelioma, five cases of sarcoma relating to hemangiopericytoma, angiosarcoma, and leiomyosarcoma, and 12 cases of primary lung cancer causing PAP.[2-16] Of all the cases of PAP arising from primary lung cancer, ten were squamous cell carcinomas, while small cell carcinoma and adenocarcinoma accounted for one case each, with all patients being males who presented with hemoptysis.[7-16] Squamous cell carcinoma of the lung is the most common neoplasm that causes PAP, due to its nature of causing necrosis, central cavitation, direct invasion, and erosion of the vessel wall.[9,11] Although PAP arising from primary lung malignancy has been previously described, its association with secondary metastasis, especially from primary breast cancer, is thought to be extremely rare. To the best of our knowledge, our case is the first reported. Only two cases of lung metastasis from a primary sarcoma causing PAP have been described.[4,7]

Most PAPs, including our case, occur peripherally, with 83% found in segmental or subsegmental branches.[7] Only one published case revealed central PAP, which was secondary to advanced lung cancer.[10] In addition, PAP typically presents as a solitary lesion.[7] However, multiple PAPs have been associated with endocarditis and metastatic sarcoma.[7]

PAP may be first detected on chest radiography, with nonspecific findings such as dense pulmonary infiltrate with hazy margins, focal lung consolidation, or a solitary well-circumscribed pulmonary nodule.[17,21] Occasionally, a PAP may not be identifiable on chest radiography due to its size, location within the lung, proximity to the mediastinum, or poor image quality.[22]

While most contrast-enhanced CT sequences may determine the extent and allow the detection of a PAP, CT angiography (CTA), in particular, CTPA protocol remains a superior imaging phase, especially for the characterization of the pseudoaneurysm and evaluating the anatomy of the pathological vessel and surrounding tissues.[7,14,17,22] Typical appearance of a PAP on contrast-enhanced CT is described as a centrally enhancing, rounded pulmonary nodule or mass with an adjacent vessel which is isodense to the central pulmonary artery.[23] Nonspecific unenhanced CT findings include ground-glass halo, consolidation, and pleural effusion.[7]

(Continue on page 30...)

(Letter to the editor continue from page 26...)

CTA will often reveal a collection of contrast outside of the blood vessel, continuous with its lumen.[22] Limitations of CT angiograms include underestimating the size of aneurysm due to the slow filling of the aneurysm or intra-aneurysmal thrombus.[14] Ultimately, catheter angiogram remains the gold standard for diagnosis and allows for interventional options such as embolization.

Symptoms associated with PAP differ depending on site, size, and the onset of conditions.[23] Chronic PAP is usually silent in its early phase; as it enlarges, a patient may experience chest pain, cough, recurrent pneumonia, shortness of breath, and hemoptysis.[23] Severe or catastrophic hemoptysis may occur when the malignant PAP is directly communicating with bronchi, creating a fistulous communication.[14]

Treatment options for PAP can be divided into conservative, surgical, and endovascular.[16] Spontaneous resolution of a small PAP has been reported, especially if it is under low pressure.[24] However, large, high-pressure PAPs, or patients presenting with severe life-threatening hemoptysis will require urgent intervention. Endovascular management includes percutaneous embolization, transcatheter embolization, and stent placement.[16,22,25,26] Various agents used for endovascular embolization have been described in the literature, such as coils, detachable balloons, patent ductus arteriosus closure devices, vascular plugs, absorbable gelatin sponges, acrylic glues, ethylene vinyl alcohol copolymer, and thrombin.[10,16,25,27-32] Different embolic agents are used depending on the size, location, neck width, and cause of the PAP.[16] Surgical management includes pulmonary artery ligation, resection of both the pseudoaneurysm and affected lung parenchyma, lobectomy, and pneumonectomy.[22] Percutaneous and transcatheter embolization

(Continue on page 41...)

(Letter to the editor continue from page 30...)

is relatively safe, cost-effective, and less invasive compared to open thoracotomy with resection.[8,17,33] Surgical management is less favorable because patients with PAP typically have less reserve and are likely to have a poor preoperative pulmonary function. Hence, lung resection is avoided whenever feasible to preserve as much functional lung parenchyma as possible.[7]

In our case, the patient's symptoms were likely attributed to the progression of chest disease and pleural effusions, with the PAP an incidental CT finding. Minimally invasive intervention may have been considered had the patient presented with hemoptysis. However, surgical intervention would have been deemed unsuitable due to the patient's advanced disease and poor pulmonary reserve.

PAP is often overlooked by reporting radiologists due to its low prevalence. A review published by Chen *et al*. revealed

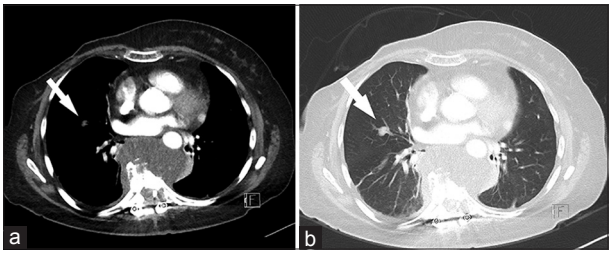

Figure 1: (a and b) Computed tomography findings of the pulmonary metastasis 2 months before occurrence of the pulmonary artery pseudoaneurysm (1 cm pulmonary nodule in the right middle lobe in proximity to right middle lobe lobar pulmonary artery)

that nearly half of all PAP cases were missed.[7] Although the prevalence of PAP in patients who present with hemoptysis is only 5%–11%, it should always be considered as a potential underlying cause, especially as prompt diagnosis and treatment

(Continue on page 45...)

(Letter to the editor continue from page 41...)

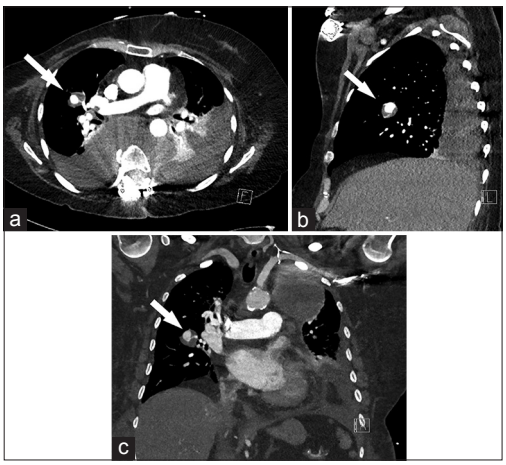

Figure 2: (a-c) Computed tomography pulmonary angiogram findings after occurrence of the pulmonary artery pseudoaneurysm (axial, sagittal, and coronal views of central arterial enhancement within the now enlarged right middle lobe metastatic deposit, with a connecting stem to the medial segmental branch of the right middle lobe pulmonary artery)

can reduce mortality related to rupture and subsequent life-threatening hemoptysis.[7]

We have presented the first reported case of PAP secondary to metastatic breast cancer. CTPA and catheter angiography are the gold standards for diagnosing PAP, with endovascular intervention the preferable treatment for suitable candidates. Clinicians and radiologists should maintain a high index of suspicion for this potentially fatal disease process. Understanding the key role that radiology plays in the diagnosis and treatment of the disease may reduce significant patient morbidity and mortality.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Pey Ling Shum[1,2], Brian Ngo[3], Xiao Chen[2,3], Robert Jarvis[2]

[1]Jeffrey Cheah School of Medicine and Health Sciences, Monash University Malaysia, Johor Bahru, Johor Malaysia, [2]Department of Radiology, Bendigo Health, Bendigo VIC, [3]Department of Radiology, Austin Health, Heidelberg VIC, Australia

Correspondence to: Pey Ling Shum,
E-mail: peylingshum@gmail.com

References

1. Nguyen ET, Silva CI, Seely JM, Chong S, Lee KS, Müller NL, *et al.* Pulmonary artery aneurysms and pseudoaneurysms in adults: Findings at CT and radiography. AJR Am J Roentgenol 2007;188:W126-34.
2. Wu XN, Chen MJ, Li DQ, Hu JG, Yu FL. Pulmonary artery pseudoaneurysm caused by a rare vascular tumor: Epithelioid hemangioendothelioma.

(Continue on page 54...)

(Letter to the editor continue from page 45...)

Thorac Cardiovasc Surg 2014;62:92-4.
3. Nakamura Y, Nishiya Y, Kawada M, Ishikawa T, Kaseno K, Fujimura M, *et al.* Primary hemangiopericytoma of the heart associated with pseudoaneurysm of the pulmonary artery – A case report. Angiology 1987;38:788-92.
4. Agarwal PP, Dennie CJ, Matzinger FR, Peterson RA, Seely JM. Pulmonary artery pseudoaneurysm secondary to metastatic angiosarcoma. Thora×2006;61:366.
5. Koch A, Mechtersheimer G, Tochtermann U, Karck M. Ruptured pseudoaneurysm of the pulmonary artery – Rare manifestation of a primary pulmonary artery sarcoma. Interact Cardiovasc Thorac Surg 2010; 10: 120-1.
6. Ablett MJ, Elliott ST, Mitchell L. Case report: Pulmonary leiomyosarcoma presenting as a pseudoaneurysm. Clin Radiol 1998;53:851-2.
7. 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;208:84-91.
8. Akpinar E, Turkbey B, Canyigit M, Peynircioglu B, Hazirolan T, Pamuk AG, *et al.* Bleeding pulmonary artery pseudoaneurysm secondary to squamous cell lung cancer: Computed tomography findings and endovascular management. Acta Radiol 2006;47:944-6.
9. Camargo Jde J, Camargo SM, Machuca TN, Bello RM. Large pulmonary artery pseudoaneurysm due to lung carcinoma: Pulmonary artery pseudoaneurysm. J Thorac Imaging 2010;25:W4-5.
10. Zhang J, Jiang S. Massive haemoptysis from a central pulmonary arterial pseudoaneurysm secondary to advanced lung cancer: Successful treatment by guglielmi detachable coil embolization. Clin Respir J 2017; 11:258-62.
11. Kim SY, Kim HR, Song JS, Hwang KE, Shin JH, Shin SN, *et al.* Pseudoaneurysm due to squamous cell carcinoma of the lung: Two cases of spontaneous resolution after chemotherapy. Cancer Res Treat 2009;41:237-40.
12. Padrones SS, Lisbona RL, Gratacos AR, Rodriguez AN, Diaz-Jimenez JP. Pulmonary artery pseudoaneurysm arising from a lung tumor. J Bronchology Interv Pulmonol 2009; 16:274-6.
13. Gomez-Jorge J, Mitchell SE. Embolization of a pulmonary artery pseudoaneurysm due to squamous cell carcinoma of the lung. J Vasc Interv Radiol 1999; 10: 1127-30.
14. Oliver TB, Stevenson AJ, Gillespie IN. Pulmonary artery pseudoaneurysm due to bronchial carcinoma. Br J Radiol 1997;70:950-1.
15. Hoffmann RT, Spelsberg F, Reiser MF. Lung bleeding caused by tumoral infiltration into the pulmonary artery – Minimally invasive repair using microcoils. Cardiovasc Intervent Radiol 2007; 30: 1282-5.
16. Park A, Cwikiel W. Endovascular treatment of a pulmonary artery pseudoaneurysm with a stent graft: Report of two cases. Acta Radiol 2007;48:45-7.
17. Lafita V, Borge MA, Demos TC. Pulmonary artery pseudoaneurysm: Etiology, presentation, diagnosis, and treatment. Semin Intervent Radiol 2007;24: 119-23.
18. Yamakado K, Takaki H, Takao M, Murashima S, Kodama H, Kashima M, *et al.* Massive hemoptysis from pulmonary artery pseudoaneurysm caused by lung radiofrequency ablation: Successful treatment by coil embolization. Cardiovasc Intervent Radiol 2010;33:410-2.
19. Sakurai J, Mimura H, Gobara H, Hiraki T, Kanazawa S. Pulmonary artery pseudoaneurysm related to radiofrequency ablation of lung tumor. Cardiovasc Intervent Radiol 2010;33:413-6.
20. Restrepo CS, Carswell AP. Aneurysms and pseudoaneurysms of the pulmonary vasculature. Semin Ultrasound CT MR 2012;33:552-66.
21. Dieden JD, Friloux LA 3rd, Renner JW. Pulmonary artery false aneurysms secondary to Swan-Ganz pulmonary artery catheters. AJR Am J Roentgenol 1987; 149:901-6.
22. Poplausky MR, Rozenblit G, Rundback JH, Crea G, Maddineni S, Leonardo R, *et al.* Swan-Ganz catheter-induced pulmonary artery pseudoaneurysm formation: Three case reports and a review of the literature. Chest 2001; 120:2105-11.
23. Kim JH, Han SH. A pulmonary artery pseudoaneurysm caused by concurrent chemoradiation therapy for lung cancer. Pak J Med Sci 2015;31:220-2.
24. 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.
25. Ghaye B, Dondelinger RF. Imaging guided thoracic interventions. Eur Respir J 2001; 17:507-28.
26. Chou MC, Liang HL, Pan HB, Yang CF. Percutaneous stent-graft repair of a mycotic pulmonary artery pseudoaneurysm. Cardiovasc Intervent Radiol 2006;29:890-2.
27. Markowitz DM, Hughes SH, Shaw C, Denny DF Jr., Wilkinson LA, White RI Jr., *et al.* Transcatheter detachable balloon embolotherapy for catheter-induced pulmonary artery pseudoaneurysm. J Thorac Imaging 1991;6:75-8.
28. Jagia P, Sharma S, Juneja R, Guleria R. Transcatheter treatment of pulmonary artery pseudoaneurysm using a PDA closure device. Diagn Interv Radiol 2011; 17:92-4.
29. Vaidyanathan B, Kannan BR, Kumar RK. Images in cardiovascular medicine. Catheter closure of pseudoaneurysm of the main pulmonary artery. Circulation 2004; 110:e322-3.
30. Chellamuthu S, Bottomley JR. Treatment of a large pulmonary artery pseudoaneurysm secondary to fungal infection using Amplatzer plugs: New embolisation devices for the management of haemoptysis. Respir Med Case Rep 2012;5:78-80.
31. Khalil A, Parrot A, Fartoukh M, Djibre M, Tassart M, Carette MF, *et al.* Pulmonary artery occlusion with ethylene vinyl alcohol copolymer in patients with hemoptysis: Initial experience in 12 cases. AJR Am J Roentgenol 2012; 198:207-12.
32. Dobies DR, Cohoon AL, Bates AA. Images in cardiovascular medicine. Repair of a perforated pulmonary artery due to a Swan-Ganz catheter using thrombin injection. Circulation 2009; 119:e521-2.
33. Pelage JP, El Hajjam M, Lagrange C, Chinet T, Vieillard-Baron A, Chagnon S, *et al.* Pulmonary artery interventions: An overview. Radiographics 2005;25: 1653-67.

How to cite this article: Shum PL, Ngo B, Chen X, Jarvis R. Pulmonary artery pseudoaneurysm secondary to metastatic breast cancer. South Asian J Cancer 2018;7:20-54.