



# Multiple Pulmonary Artery Pseudoaneurysms Secondary to Metastatic Uterine Leiomyosarcoma

CASE REPORT

TEETAWAT JEARSIRIKUL 

SURAJAI JUNNHU 

NANTAKA KIRANANTAWAT 

\*Author affiliations can be found in the back matter of this article

]u[ubiquity press

## ABSTRACT

**Teaching point:** The probable mechanism of pseudoaneurysm formation related to metastatic neoplasm is a tumor embolus penetrating and destroying the vessel wall.

Pulmonary artery pseudoaneurysm (PAP) related to metastatic neoplasm is rare. We describe a unique case of multiple PAPs secondary to metastatic uterine leiomyosarcoma and demonstrate the serial chest computed tomography to support the theory that the tumor begins as a tumor embolus, followed by infiltration and breakdown of the vessel wall, leading to aneurysmal dilatation and invading the perivascular tissue.

CORRESPONDING AUTHOR:

**Nantaka Kiranantawat**

Prince of Songkla University, TH

[Nantaka.k@psu.ac.th](mailto:Nantaka.k@psu.ac.th)

KEYWORDS:

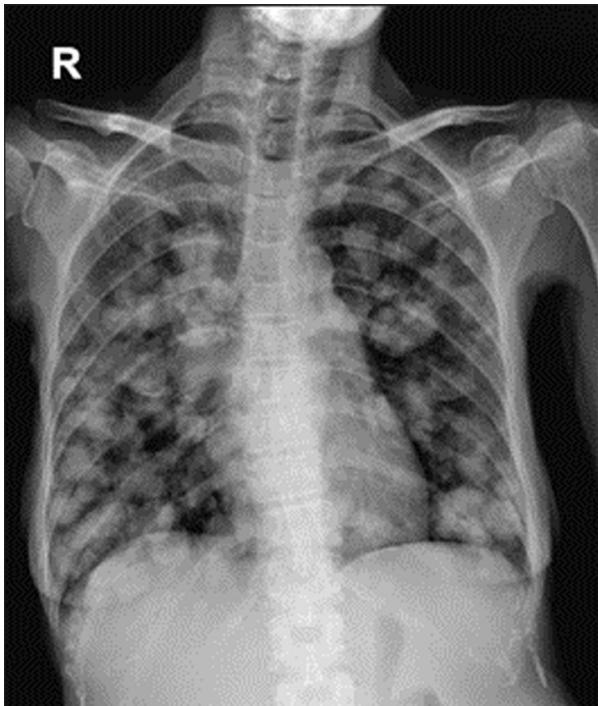
Pulmonary arterial pseudoaneurysm; metastasis; leiomyosarcoma; sarcoma; pulmonary metastasis

TO CITE THIS ARTICLE:

Jearsirikul T, Junnhu S, Kiranantawat N. Multiple Pulmonary Artery Pseudoaneurysms Secondary to Metastatic Uterine Leiomyosarcoma. *Journal of the Belgian Society of Radiology*. 2021; 105(1): 52, 1-4. DOI: <https://doi.org/10.5334/jbsr.2561>

## CASE REPORT

A 49-year-old female with known uterine leiomyosarcoma and lung metastasis with multiple tumor thrombi in pulmonary arteries was receiving palliative chemotherapy. Prior to her fourth chemotherapy session, the patient developed acute-onset dyspnea. Chest radiograph revealed a larger size of the multiple well-defined pulmonary masses scattered in both lungs; therefore, progressive pulmonary metastasis was suspected (*Figure 1*). Computed tomography pulmonary angiography (CTPA) was performed to rule out acute pulmonary thromboembolism and showed innumerable



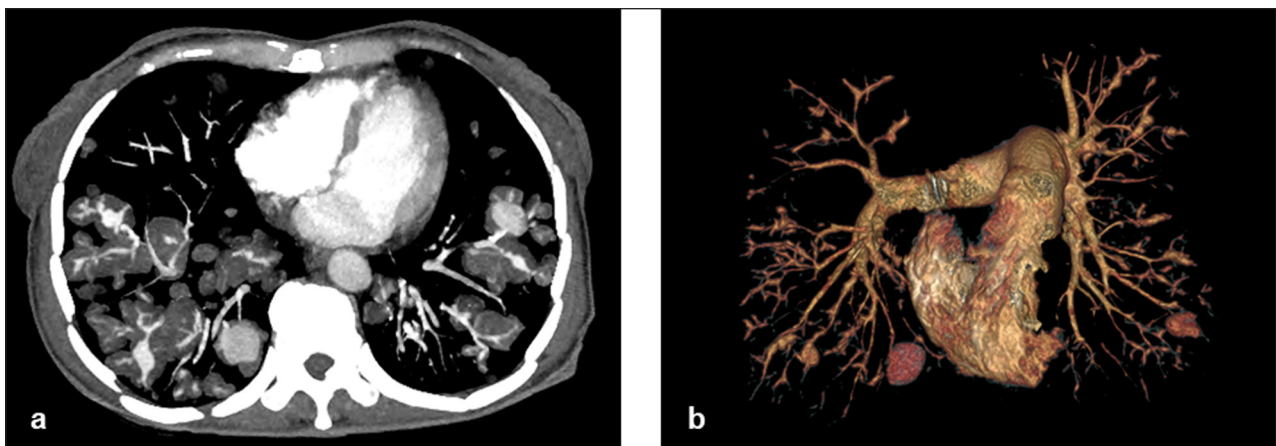
**Figure 1** Chest radiograph showing multiple, round, variably sized masses scattering in both lungs, compatible with pulmonary metastasis.

heterogeneously enhanced masses scattered in both lungs, surrounding segmental branches of pulmonary arteries. Multiple fusiform pulmonary artery pseudoaneurysms surrounded by soft tissue masses were also noted (*Figure 2*). A review of serial chest computed tomography (CT) revealed tumor emboli in the peripheral branches of pulmonary arteries (*Figure 3a*), which finally developed into pseudoaneurysms with surrounding soft tissue masses (*Figure 3b–3f*).

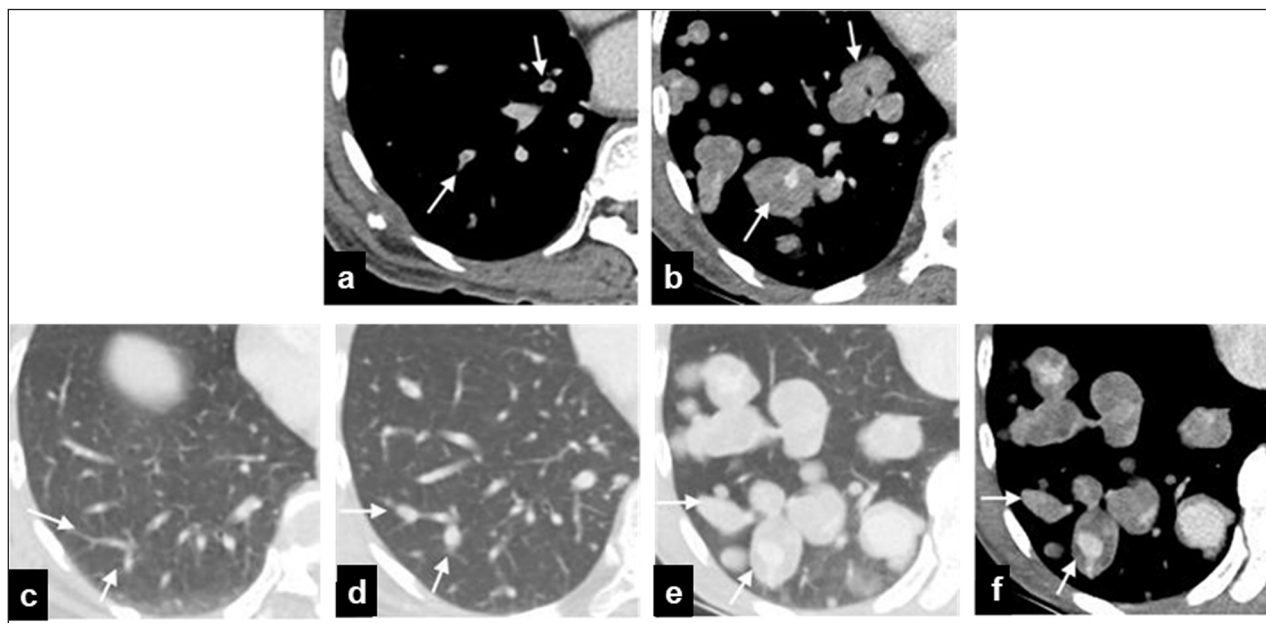
## DISCUSSION

PAP is defined as a focal dilatation of the pulmonary artery that does not comprise all three layers of the vessel wall. PAP itself is a rare condition, usually arising secondary to infection. Malignancy is a rare cause of PAP with a predilection towards primary cancer of the lungs [1]. From previous literature, we found that PAP was related to metastatic neoplasm in five cases, including metastatic sarcoma, synovial sarcoma of the right atrium, angiosarcoma of soft tissue in the neck, atrial myxoma, and breast cancer [2–6]. Only two cases revealed multiple PAPs: metastatic sarcoma and atrial myxoma [2, 6].

The mechanism of PAP secondary to metastatic neoplasm could be explained by the “metastasize and infiltrate” theory that described the formation of cerebral artery aneurysms due to metastatic cardiac myxoma [7]. The theory proposes that the process begins as a tumor embolus, followed by infiltration and occupation of the subintimal layer of vessel walls, causing loss of the normal vessel integrity, and lastly forming a pseudoaneurysm. This theory was also supported histologically by two case reports of an intracerebral pseudoaneurysm, and multiple bilateral PAPs due to cardiac myxoma [5, 8]. The authors also demonstrated tumor cells invading through the vessel walls into surrounding alveolar tissue [5].



**Figure 2** (a) Axial MIP image showing multiple masses, scattering in both lungs and surrounding segmental/subsegmental branches of pulmonary arteries, and multiple PAPs. (b) Coronal 3D Volume Rendered image showing multiple PAPs scattering in both lungs. The two largest ones are located in both lower lobes.



**Figure 3** (a) and (b) Serial axial chest CT within one month showing aneurysmal dilatation and developing soft tissue masses surrounding the previously seen tumor emboli in the peripheral branches of right pulmonary arteries (arrows). (c–f) Serial axial chest CT at eight months prior (c), one month prior (d), and the present study (e) and (f) showing gradual dilatation of peripheral branches of right pulmonary arteries, and finally pseudoaneurysms with surrounding soft tissue masses (arrows).

A similar mechanism of pathogenesis is thought to have occurred in this case. From the serial chest CT, the PAPs with surrounding soft tissue masses had previously shown intraluminal thrombus and dilated pulmonary artery, which resembled tumor thrombus (Figure 3a and 3b). The serial chest CT also showed gradual dilatation of peripheral branches of right pulmonary arteries (Figure 3c–3f); this could be due to tumor growth and continued destruction of vessel walls.


Abnormal dilatation of the pulmonary arteries and veins was also observed in the patient's CTPA with similar findings previously described in a reported case of metastatic uterine sarcoma [9]. The only difference is that there was a clear connection between the dilated vessels with the pulmonary circulation in this patient, unlike the reported case in which the dilated vessel occurred in isolation from the pulmonary circulation, thought to have been formed by the mass itself.

In conclusion, PAPs caused by metastatic neoplasm may firstly begin as tumor emboli, later progressing into pseudoaneurysms as we elucidated in the serial chest CTs and CTPA findings in this case.

## COMPETING INTERESTS

The authors have no competing interests to declare.

## AUTHOR AFFILIATIONS

**Teetawat Jearsirikul**  [orcid.org/0000-0002-8460-8640](https://orcid.org/0000-0002-8460-8640)  
Prince of Songkla University, TH

**Surajai Junnhu**  [orcid.org/0000-0002-4911-0425](https://orcid.org/0000-0002-4911-0425)

Prince of Songkla University, TH

**Nantaka Kiranantawat**  [orcid.org/0000-0003-0828-8486](https://orcid.org/0000-0003-0828-8486)

Prince of Songkla University, TH

## REFERENCES

- Gupta M, Agrawal A, Iakovou A, Cohen S, Shah R, Talwar A.** Pulmonary artery aneurysm: A review. *Pulm Circ.* 2020; 10(1): 2045894020908780. DOI: <https://doi.org/10.1177/2045894020908780>
- Chen Y, Gilman MD, Humphrey KL, et al.** Pulmonary artery pseudoaneurysms: Clinical features and CT findings. *AJR Am J Roentgenol.* 2017; 208: 84–91. DOI: <https://doi.org/10.2214/AJR.16.16312>
- Bean SH, Sadler DJ, Gray RR, So CB, Morris D.** Embolization of pulmonary pseudoaneurysms secondary to metastatic synovial sarcoma of the right atrium. *J Vasc Interv Radiol.* 1999; 10(5): 649–52. DOI: [https://doi.org/10.1016/S1051-0443\(99\)70096-5](https://doi.org/10.1016/S1051-0443(99)70096-5)
- Agarwal PP, Dennie CJ, Matzinger FR, Peterson RA, Seely JM.** Pulmonary artery pseudoaneurysm secondary to metastatic angiosarcoma. *Thorax.* 2006; 61: 366. DOI: <https://doi.org/10.1136/thx.2005.048645>
- Rokadia HK, Heresi GA, Tan CD, Raymond DP, Budd GT, Farver C.** A 33-year-old man with multiple bilateral pulmonary pseudoaneurysms. *Chest.* 2015; 148(4): e112–e117. DOI: <https://doi.org/10.1378/chest.15-0624>
- Shum PL, Ngo B, Chen X, Jarvis R.** Pulmonary artery pseudoaneurysm secondary to metastatic breast cancer. *South Asian J Cancer.* 2018; 7(1): 20. DOI: [https://doi.org/10.4103/sajc.sajc\\_157\\_17](https://doi.org/10.4103/sajc.sajc_157_17)

7. **New PF, Price DL, Carter B.** Cerebral angiography in cardiac myxoma. Correlation of angiographic and histopathological findings. *Radiology*. 1970; 96(2): 335–45. DOI: <https://doi.org/10.1148/96.2.335>
8. **Furuya K, Sasaki T, Yoshimoto Y, Okada Y, Fujimaki T, Kirino T.** Histologically verified cerebral aneurysm formation secondary to embolism from cardiac myxoma. *Case report. J Neurosurg*. 1995; 86(1): 170–3. doi: <https://doi.org/10.3171/jns.1995.83.1.0170>
9. **Jang HJ, Kim SS, Park HS, Lee JE, Kim JH.** Pulmonary metastasis originated from uterine sarcoma presenting as multiple nodules with tortuous serpentine aneurysmal dilated intratumoral vessels: A case report. *J Korean Soc Radiol*. 2018; 78(4): 284–288. DOI: <https://doi.org/10.3348/jksr.2018.78.4.284>

---

**TO CITE THIS ARTICLE:**

Jearsirikul T, Junnhu S, Kiranantawat N. Multiple Pulmonary Artery Pseudoaneurysms Secondary to Metastatic Uterine Leiomyosarcoma. *Journal of the Belgian Society of Radiology*. 2021; 105(1): 52, 1–4. DOI: <https://doi.org/10.5334/jbsr.2561>

Submitted: 15 June 2021   Accepted: 06 September 2021   Published: 23 September 2021

**COPYRIGHT:**

© 2021 The Author(s). This is an open-access article distributed under the terms of the Creative Commons Attribution 4.0 International License (CC-BY 4.0), which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited. See <http://creativecommons.org/licenses/by/4.0/>.

*Journal of the Belgian Society of Radiology* is a peer-reviewed open access journal published by Ubiquity Press.