

Bilateral pulmonary nodules: A case of meningotheeliomatosis

Filipa Jesus¹, Joana A. Ribeiro¹, Rita Gomes², Conceição Souto Moura³, Alcina Tavares¹, Luís Ferreira¹

The differential diagnosis for diffuse bilateral pulmonary nodules is extensive and includes infections, inflammatory disorders and metastatic malignancy (Table 1)¹.

Minute pulmonary meningotheelial-like nodules (MPMN) result from epithelioid cell proliferation within the interstitium, and can present as a solitary pulmonary nodule or, less frequently, as diffuse nodules, termed diffuse pulmonary meningotheeliomatosis (DPM)².

In DPM these cells form small nodules of ground-glass appearance on chest CT scan, with/without central lucency ('cheerio sign') throughout the lung tissue and typically present as diffuse and bilateral randomly distributed nodules³. Most are too small to be detected by CT, but the autopsy incidence has been reported as 7.2%⁴.

DPM/MPMN are benign and are more common in females with age in the sixth to seventh decade. Patients are often asymptomatic or they may present with non-specific cardiorespiratory symptoms⁵. The exact etiology remains unclear, however, these nodules are more commonly associated with pulmonary thromboembolic disease or smoking-related interstitial lung disease⁶. A higher incidence of these nodules has been reported in patients with primary malignant pulmonary tumors, specifically with lung adenocarcinoma⁴. Definitive diagnosis requires lung biopsy, typically surgical³.

We report a case of diffuse pulmonary meningotheeliomatosis in a patient with a past medical history of lung adenocarcinoma.

An 81-year-old woman, non-smoker, with a past medical history of stage IA lung adenocarcinoma submitted 3 years before to superior right lobectomy, presented to pulmonary clinic for follow-up and did routine chest CT which showed multiple bilateral micronodules of ground glass appearance, with 2–5 mm, with basal predominance (Figures 1 A, B and C), raising the possibility of metastatic lesions. She denied any symptomatology associated, namely chest pain, dyspnea or systemic symptoms.

To study these lesions and evaluate the possibility of a relapse of the malignant disease, she did an 18-FDG PET scan that showed a micronodular pattern in both lungs, functionally uncharacterizable due to the reduced dimensions of the nodules, without any other lesions.

Due to dimensions of the lesions and the difficult approach through CT-guided transthoracic biopsy, a video-assisted thoracoscopic (VATS) biopsy was performed. The histopathological examination revealed that these nodules consisted of pulmonary meningotheeliomatosis, excluding metastatic lung lesions (Figures 1 D, E and F). The patient is currently asymptomatic and maintains in follow-up, without signs of malignancy in the 2 years period since the detection of the nodules.

DMP is a relatively rare condition, apparently clinically innocuous and with conservative management³. Despite this, we consider that awareness of this entity is important as it can be confused with other diseases, namely metastatic disease as in the case abovementioned.

Differential diagnosis in these cases can be difficult, especially in patients with history of a concomitant malignancy, and will require correlation with clinical and radiological findings, as well as lung biopsy for definitive diagnosis.

AFFILIATION

1 Department of Pulmonology, Unidade Local de Saúde da Guarda - E.P.E. Hospital Sousa Martins, Guarda, Portugal

2 Department of Pulmonology, Centro Hospitalar de Entre o Douro e Vouga E.P.E., Santa Maria da Feira, Portugal

3 Department of Pathology, Centro Hospitalar e Universitário Hospital de São João E.P.E., Porto, Portugal

CORRESPONDENCE TO

Filipa Jesus. Department of Pulmonology, Unidade Local de Saúde da Guarda - E.P.E. Hospital Sousa Martins, Av. Rainha D. Amélia, 6301-857 Guarda, Portugal.

E-mail: filipa.jesus@ulsguarda.min-saude.pt
ORCID ID: <https://orcid.org/0000-0002-3640-6374>

KEYWORDS

pulmonary nodules, lung adenocarcinoma, meningotheeliomatosis

Received: 14 February 2023

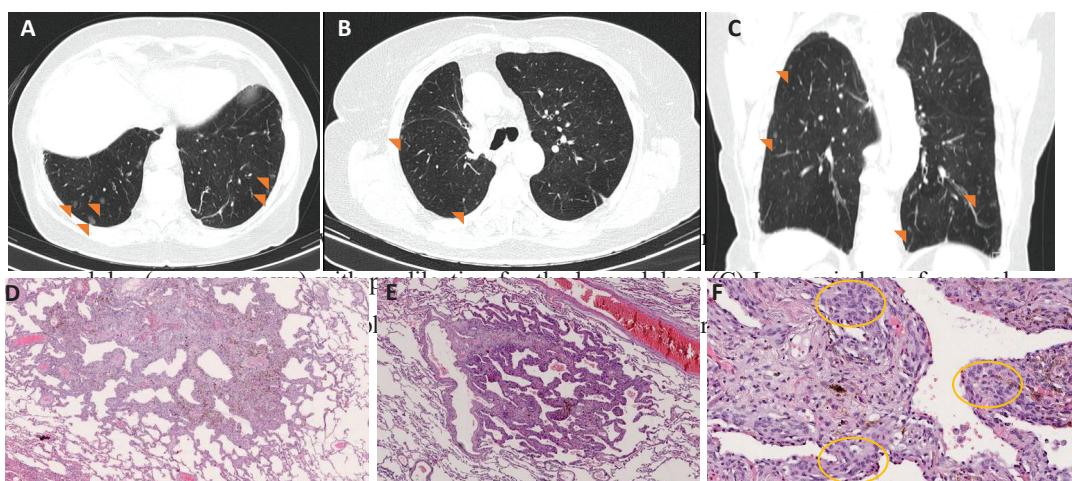
Revised: 16 March 2023

Accepted: 12 April 2023

Table 1. Differential diagnosis of diffuse pulmonary bilateral nodules

Infections	Diffuse lung diseases	Malignancies
Miliary tuberculosis	Sarcoidosis	Metastasis
Atypical (non-tuberculous) Mycobacterial infection	Hypersensitivity pneumonitis	Bronchioloalveolar carcinoma
Fungal infection	Diffuse pulmonary meningotheiomatosis	Lymphangitic carcinomatosis
Septic emboli	Pneumoconiosis (namely silicosis)	
	Amyloidosis	Kaposi sarcoma

Figure 1. (A, B) Lung window of axial chest CT scan showing multiple ground glass lung nodules (orange arrows) with predilection for the lower lobes; (C) Lung window of coronal chest CT scan showing multiple ground glass lung nodules (orange arrows); (D, E) Histopathological findings, showing areas of nodular configuration, with expansion of the alveolar septa and architectural preservation; (F) These nodular areas are constituted by polygonal cells, with uniform nucleus (orange circles), with positivity for EMA and vimentin



CONFLICTS OF INTEREST

The authors have completed and submitted the ICMJE Form for Disclosure of Potential Conflicts of Interest and none was reported.

FUNDING

There was no source of funding for this research.

ETHICAL APPROVAL AND INFORMED CONSENT

Ethical approval was not required for this study. Patient's informed consent was obtained concerning the publication of this case report.

DATA AVAILABILITY

The data supporting this research are available from the authors on reasonable request.

PROVENANCE AND PEER REVIEW

Not commissioned; externally peer reviewed.

REFERENCES

- Boitsios G, Bankier AA, Eisenberg RL. Diffuse pulmonary nodules. AJR Am J Roentgenol. 2010;194(5):W354-W366. doi:[10.2214/AJR.10.4345](https://doi.org/10.2214/AJR.10.4345)
- Suster S, Moran CA. Diffuse pulmonary meningotheiomatosis. Am J Surg Pathol. 2007;31(4):624-631. doi:[10.1097/PAS.0000213385.25042.CF](https://doi.org/10.1097/PAS.0000213385.25042.CF)
- Yun G, Huang T, O'Dwyer D, Chugtai A, Agarwal P. Diffuse pulmonary meningotheiomatosis. Clin Imaging. 2021;70:111-113. doi:[10.1016/j.clinimag.2020.10.007](https://doi.org/10.1016/j.clinimag.2020.10.007)
- Mizutani E, Tsuta K, Maeshima AM, Asamura H, Matsuno Y. Minute pulmonary meningotheelial-like nodules: clinicopathologic analysis of 121 patients. Hum Pathol. 2009;40(5):678-682. doi:[10.1016/j.humpath.2008.08.018](https://doi.org/10.1016/j.humpath.2008.08.018)
- Alkurashi AK, Almodallal Y, Albitar HAH, Cheville JC, Iyer VN. Diffuse Pulmonary Meningotheiomatosis: A Rare Lung Disease Presenting with Diffuse Ground-Glass Opacities and Cavitation. Am J Case Rep. 2020;21:e926172. doi:[10.12659/AJCR.926172](https://doi.org/10.12659/AJCR.926172)
- Mukhopadhyay S, El-Zammar OA, Katzenstein AL. Pulmonary

meningotheelial-like nodules: new insights into a common but poorly understood entity. Am J Surg Pathol. 2009;33(4):487-495. doi:[10.1097/PAS.0b013e31818b1de7](https://doi.org/10.1097/PAS.0b013e31818b1de7)