

CASE REPORT

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PET/CT imaging of giant primary pulmonary meningioma: a case report and literature review

Yawen Feng¹, Peng Wang¹, Yufei Liu² and Wenli Dai^{1*}

Abstract

Background An ectopic meningioma, such as a primary pulmonary meningioma (PPM), is a rare type of tumor that primarily originates outside of the central nervous system. The most common presentation of PPM is isolated pulmonary nodules or masses, and most of them are benign. Only sporadic cases have been reported. This case reported a giant primary pulmonary meningioma and systematically reviewed previously reported cases in the literature.

Case presentation A 55-year-old female suffered from asthma after activity, chest tightness, and a persistent dry cough for 2 months. Chest computed tomography (CT) showed a huge mass with calcification in the left lower lobe. And positron emission tomography (PET)/CT revealed mild FDG accumulation of the mass. The mass was finally surgically removed and PPM was confirmed according to histopathologic examinations.

Conclusion PPM is a rare disease with heterogeneity not only in CT features but also in glucose metabolism. FDG uptake levels do not identify benign from malignant, benign PPM may have high FDG uptake and malignant may have low.

Keywords Primary pulmonary meningioma, ¹⁸F-fluorodeoxyglucose, PET/CT, Case report

Background

Meningiomas are common tumors of the nervous system originating from arachnoid cells, which often occur intracranially or in the spinal canal, and are mostly benign. Meningiomas occurring in tissues and organs not covered by meninges are called ectopic meningiomas which are very rare tumors that commonly occur in the head

and neck or, less frequently, in the skin [1, 2]. Primary meningioma in lung tissue is defined as primary pulmonary meningioma. Since the first case report in 1982 by Kemnitz et al [3], only a small number of cases of PPMs have been reported. With the development of medical imaging disciplines, we can make a preliminary diagnosis or differential diagnosis of PPM through imaging. Herein, we report a case of a giant PPM diagnosed with ¹⁸F-FDG PET/CT imaging and a review of the literature on previous instances of ectopic meningioma of the thoracic cavity diagnosed with ¹⁸F-FDG PET/CT imaging.

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Case presentation

A 55-year-old female patient was admitted to our hospital complaining of asthma after activity, chest tightness, and a persistent dry cough for 2 months. A physical examination of the left side of the chest revealed decreased respiratory motion, decreased breath sounds, and turbid percussion in the left side of the chest. After admission, routine blood tests, lung cancer tumor markers and node antacid staining did not show any significant abnormalities. Pleural fluid cytopathological examination showed a small number of lymphocytes, macrophages, mesothelial cells, and no malignant tumor cells. A chest computed tomography (CT) performed a lobulated pulmonary mass with calcification in the left lower lobe, and adjacent pleural hypertrophy and adhesions. The patient underwent the ^{18}F -FDG PET/CT examination to clarify the diagnosis. On ^{18}F -FDG PET imaging (Fig. 1), the standardized uptake value (SUV) of the mass increased unevenly, with SUVmax from 4.4 to 8.1. The size of the mass is about 76 mm \times 70 mm \times 59 mm. And without lesions identified in other locations in his body (including nervous system and spine). The patient's diagnosis and treatment process showed in the Table 1. Due to massive calcified lesions and mild high metabolic activity, the nuclear medicine physician considered the mass to be benign. For further diagnosis and treatment, the patient received a surgery. Intraoperative exploration revealed an 8 cm diameter calcified mass between the left

lower lung and the diaphragm with adhesions to the surrounding lung tissue and the diaphragm. Subsequently, the patient underwent left thoracic adhesion, mass excision, wedge resection of the left lower lung lobe and intraoperative frozen pathological examination. Gross examination revealed a grayish-white mass measuring 9.5cm \times 8.4cm \times 5.3 cm. Microscopic examination showed mainly spindle cells, with mild cell morphology. The tumor involved the pleura with fusiform nests of cells arranged in fascicles or whorls and extensive calcification. Immunohistochemistry showed positivity for epithelial membrane antigen (EMA), progesterone receptor (PR), somatostatin receptor 2 (SSTR2), CD34, and S-100, and negativity for Desmin, STAT6, ALK(1A4), and SMA. The Ki-67 index was about 20% positive. These morphological and immunohistochemical features were suggestive of a benign PPM (WHO 1 grade) (Fig. 2).

Discussion

The incidence of ectopic meningioma is very rare (2%) and can be located in various anatomical sites such as the scalp, maxillary sinus, sinus lung, parotid gland, skin, and peripheral nerves [4]. The pathogenesis of PPM is still unclear, and it is generally believed that ectopic meningioma may derive from pluripotential subpleural mesenchymal cells or heterotopic embryonic rests of arachnoid cells [1, 2]. Most patients with PPM do not have significant symptoms, while some patients have respiratory.

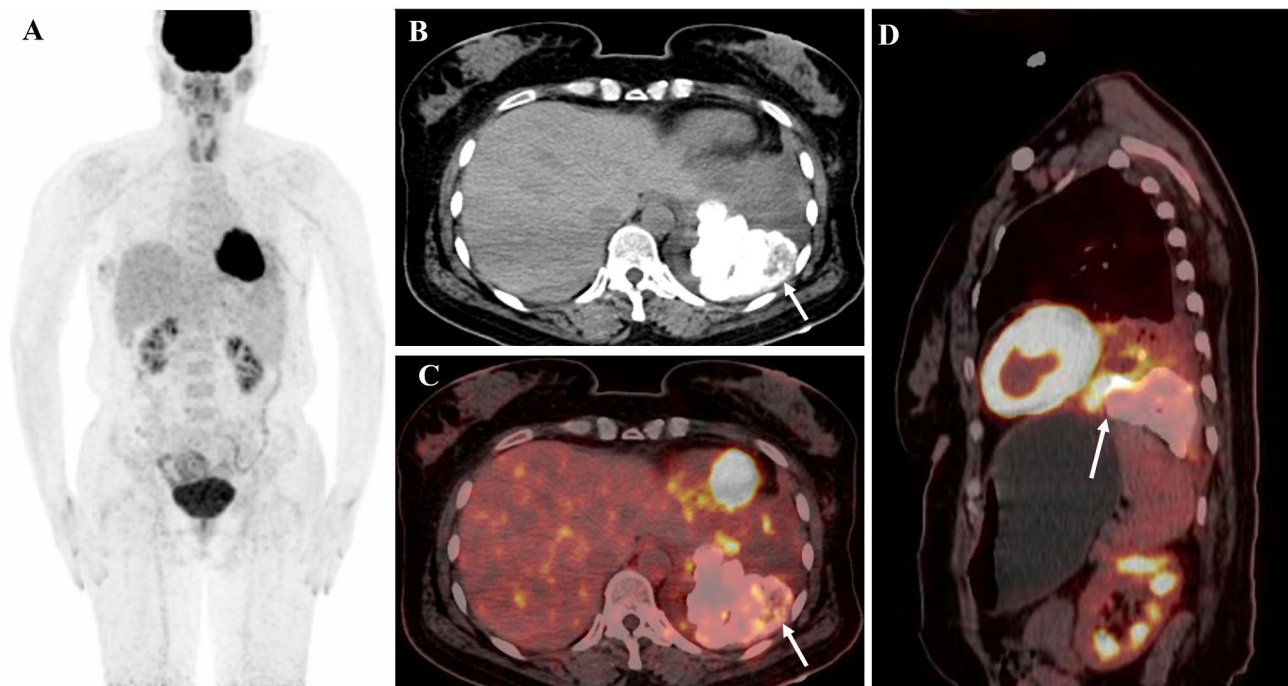


Fig. 1 The maximum intensity projection PET Image (A) in the whole body. Transverse chest CT showing a huge tumor with massive calcification (B), and fusion PET/CT (C) images demonstrating a huge tumor with mild high metabolic activity (SUVmax 4.4) and the size of it about 76 mm \times 70 mm \times 59 mm. Sagittal PET/CT image (D) demonstrating the site of the highest metabolism (SUVmax 8.1)

Table 1 Patient examination and treatment procedure

Items	Detailed information
Laboratory Examination	
Hematological	
WBC	7.27 × 10 ⁹ /L
RBC	4.16 × 10 ¹² /L
Blood platelet	284 × 10 ⁹ /L
Lung cancer tumor markers	
CEA	1.4ng/ml
NSE	14.1ng/ml
Cyfra21-1	1.2ng/ml
ProGRP	32.9pg/ml
SCC	0.3ng/ml
Examinations of pleural fluid	
TP	66.36 g/L
ALB	31.28 g/L
LDH	1823IU/L
T-SPOT	(-)
Acid-fast staining	(-)
Imaging examinations	
CT of the chest	A lobulated mass in the lower lobe of the left lung with massive calcification, measuring about 76 mm × 70 mm × 59 mm.
¹⁸ F-FDG PET/CT	Uneven metabolism of the masses, with SUVmax from 4.4 to 8.1.
Treatment	Surgical operation
Pathological results	Meningioma (Benign, WHO I Grade)
Follow up	No recurrent lesions were found in the operated area at the 1-year postoperative review CT.

WBC: white blood cell; RBC: red blood cell; CEA: carcinoembryonic antigen; NSE: euron specific enolase; Cyfra21-1: cytokeratin 19 fragment; ProGRP: pro-gastrin-releasing peptide; SCC: squamous cell carcinoma; TP: Total protein; ALB: albumin; CT: computed tomography; ¹⁸ F-FDG PET: 18 F-fluorodeoxyglucose positron emission tomography.

Clinical symptoms may be related to the position of the lesion and the size of the lesion [5]. In this case, the patient had chest tightness, shortness of breath, and cough, probably due to the huge mass adjacent to the pleura. Most PPMs were benign, and only a few cases are malignant [6–10]. Patients with benign PPM have a better prognosis and generally do not recur or metastasize distantly.

Imaging characteristics can be distinguished to some degree from other types of tumors. On chest CT scans, PPMs usually appear as isolated, round, solid, well-defined nodules or masses, with or without calcification. But a few cases present with ground-glass nodules or multiple cystic lesions [11, 12]. The size of benign PPMs ranged from 0.4 to 6 cm in diameter (median: 2 cm). The size of malignant PPMs ranged from 1.5 to 15 cm in diameter (median: 6.4 cm) [5]. Although the average diameter of malignant PPMs is longer than benign PPMs, it is not possible to distinguish between benign and malignant based on the size of the tumor alone. As we

reported a huge PPM, measuring 9.5cmx8.4cmx5.3 cm, but the pathology results confirmed that it was a benign tumor.

To further learn the characteristics of ¹⁸ F-FDG PET/CT images of PPMs, we performed a systematic search of PubMed and web of science using the keywords “primary pulmonary meningioma”, “PET”, “ectopic meningioma”. And finally, we got 11 case reports about primary pulmonary meningioma with complete data of PET/CT and listed them in Table 2. On ¹⁸ F-FDG PET, PPMs exhibit mild to high metabolic activity, and the average value of SUVmax is 4.36, ranging from 0.6 to 12.9. The lesion with the SUVmax of 0.6 was a well-defined nodular lesion, measuring approximately 1.3 cm × 1.3 cm × 1.5 cm, whose pathological findings were benign PPM [13]. The lesion with SUVmax of 12.9 was also a benign PPM, measuring 1.5 cm, and appeared as a lobulated nodule on CT images [14]. The largest PPM was the one we reported in this article. The size was 9.5 cm x 8.4 cm x 5.3 cm and the SUVmax value was from 4.4 to 6.2, showing mild to moderate uptake of ¹⁸ F-FDG. Due to the mass having a multitude of calcified lesions and mostly mild FDG uptake, the mass was diagnosed as a benign lesion at the time of initial diagnosis. Therefore, the level of SUVmax alone cannot evaluate the nature of PPMs (benign vs. malignant). The case reported by Andrea Cimini et al [7] also confirmed this view. In their case report, a patient had two PPMs, and the malignant PPM had lower SUVmax values than the benign PPM. Consequently, PPMs are mainly diagnosed by pathology, while imaging medicine can initially distinguish PPMs from other tumors.

Benign PPMs can be cured by surgical treatment. Malignant PPMs are prone to recurrence or metastasis hence the prognosis is poor. In general, most PPM have a good prognosis without recurrence or metastasis [16], and the main strategy of treatment is surgical resection [21]. The patient, in our case, received surgery and had been followed up for 9 months without tumor recurrence.

Conclusion

In this paper, we reported a huge mass in the, with massive calcified lesions, and mild to moderate metabolism, the final diagnosis was benign PPM. We reviewed the literature and found that most of PPMs usually appear as isolated, round, solid, well-defined nodules or masses, with or without calcification on CT scan and exhibit mild to high metabolic activity on PET scan, and the level of SUVmax alone cannot evaluate the nature of PPMs (benign vs. malignant).

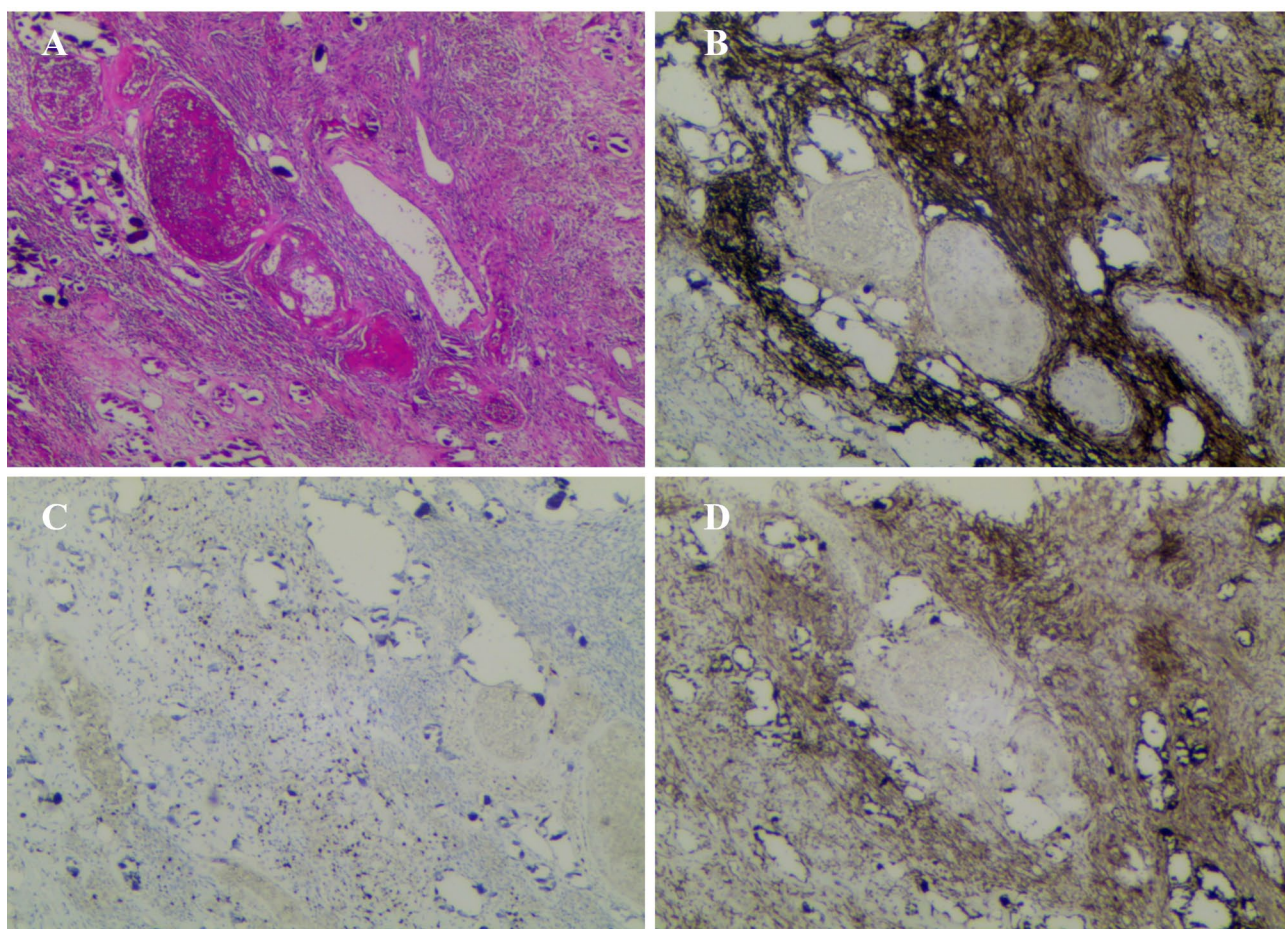


Fig. 2 Pathological examination. Hematoxylin-eosin Stain (A), Original Magnification 100x. The tumor cells stained positive for EMA (B, 100x) and SSTR2 (D, 100x), and Ki-67 index was about 20% (C, 100x). These findings are supporting the diagnosis of pulmonary meningioma

Table 2 Imaging and PET/CT features of thoracic ectopic meningioma case

NO.	Ref.	Age	gender	Symptom	CT feature	Site	Tumor size (cm)	Contrast-enhanced CT	PET/CT	His-tology
1	Danbin Zhang [5]	47	F	None	A lobulated mass with calcification.	LL	6.9×5.7×6.1	Mild enhancement	SUVmax 4.4	B
2	Maoqing Jiang [13]	70	M	None	A solitary pulmonary nodule with well-defined.	RL	1.3×1.3×1.5	Mild enhancement	SUVmax 0.6	B
3	CURA [15]	58	F	None	A well-circumscribed mass.	RU	2	N	Metabolic activity	B
4	Meirelles [14]	48	M	None	A lobulated nodule.	RU	1.5	N	SUVmax 12.9	B
5	Incarbone [16]	24	M	None	A solitary pulmonary nodule.	RU	1.4	Mild enhancement	SUVmax 10.14	B
6	Lepanto [17]	60	F	None	A solitary pulmonary nodule.	LU	1.2	N	SUVmax 1.2	B
7	Bae [18]	43	F	None	A solitary pulmonary nodule.	LU	1.7	Well enhancement	SUVmax 2.48	B
8	Gürçay [12]	55	F	Coughing	A solitary ground-glass nodule.	RU	2×2×1.8	N	SUVmax 1.89	B
9	Oh [19]	54	M	None	Multiple lung nodules	RU LU	0.96 1.25	N N	SUVmax 3.1 SUVmax 2.3	B B
10	Cimini [7]	80	M	None	A solitary pulmonary nodule. A solitary pulmonary nodule.	RU LR	1.4 1.2	N Well enhancement	SUVmax 4.63 SUVmax 2.46	B M
11	BAŞ [20]	57	M	Coughing	A solitary pulmonary nodule.	LL	1.0	N	¹⁸ F-FDG no uptake	B
12	This report	55	F	Asthma, cough, chest tightness	A lobulated mass with calcification.	LL	9.5×8.4×5.3	N	SUVmax 6.2	B

F: Female; M: Male; None: No symptoms; B: Benign; M: Malignant; RL: Right lower lobe; RU: Right upper lobe; LL: Left lower lobe; LU: Left upper lobe; N: Not reported.

Abbreviations

¹⁸ F-FDG-PET/CT	18 F-fluorodeoxyglucose positron-emission tomography-computed tomography
SUVmax	Maximum Standardized Uptake Value
PPM	Primary pulmonary meningioma
CT	computed tomography
F	Female
M	Male
None	No symptoms
B	Benign
M	Malignant
RL	Right lower lobe
RU	Right upper lobe
LL	Left lower lobe
LU	Left upper lobe
N	Not reported.

Acknowledgements

Not applicable.

Author contributions

Yawen Feng drafted the manuscript and Peng wang, Wenli Dai participated in the revision of the manuscript for important intellectual content. Yufei Liu made pathological sections and provided pathological pictures. All authors read and approved the final manuscript.

Funding

We received neither funding nor sponsorship for this study nor any publication of this article.

Data availability

Data sharing is not applicable to this article as no datasets were generated or analysed.

Declarations

Ethics approval and consent to participate

Written informed consent was obtained from the patient to participate in this case report.

Consent for publication

Written informed consent for publication of clinical details and clinical images was obtained from the patient. A copy of the consent form is available for review by the Editor of the journal.

Competing interests

The authors declare no competing interests.

Received: 27 November 2022 / Accepted: 5 April 2023

Published online: 03 May 2023

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