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CASE REPORT

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Primary pulmonary meningioma presenting as multiple lung nodules: A case report

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Abstract

Primary pulmonary meningioma (PPM) is a very rare and mostly benign disease. Although PPM is incidentally detected as a solitary pulmonary nodule on X-ray or chest computed tomography, it does not have unique radiological characteristics; thus, it is difficult to differentiate it from other lung tumors. A healthy 54-year-old man visited our hospital because of multiple variable-sized lung nodules. He had no contributive medical history and no abnormality that was suggestive of extrapulmonary malignancy. Video-assisted thoracoscopic wedge resection was undertaken for diagnosis, and the tumor cells were histopathologically confirmed as PPM. Brain magnetic resonance imaging revealed no intracranial tumor. The patient has been well and without any progression of the remaining lesions over 24 months. Here, we present the clinicopathological features of this case in which the patient's nodules were mistaken for multiple metastatic lung nodules.

KEYWORDS

immunohistochemistry, meningioma, multiple pulmonary nodules

INTRODUCTION

Meningioma is the most common intracranial tumor, accounting for approximately 20% of brain tumors.[1] However, extracranial primary meningioma is very rare. Ectopic meningiomas, which mainly originate from outside of the central nervous system, account for approximately 2% of meningiomas.[2] Among them, primary pulmonary meningioma (PPM) is extremely rare, and there has been less than 60 cases reported in the literature internationally since the first case in 1982.[3,4] Here, we report the clinicopathological features of PPM, which is often mistaken for multiple metastatic lung nodules in a 54-year-old male.

CASE REPORT

A 54-year-old man was referred for further evaluation of multiple lung nodules found on chest computed tomography (CT) during a routine health check-up. He had a smoking history of 60 pack-years, and his medical history was not contributive. He did not complain about any respiratory symptoms. There was no remarkable finding on physical examination. Initial contrast-enhanced CT revealed several variable-sized nodules on both lungs including 10 in the left and seven in the right lung, which were suspected to be metastases from extra-thoracic malignancy (Figure 1). Abdominopelvic CT, endoscopy of the gastrointestinal tract, ultrasonography of the prostate, and blood tumor markers, including prostate-specific antigen, carbohydrate antigen 19–9, and carcinoembryonic antigen were performed to identify the primary site of the tumor; however, there were no abnormal findings that were suggestive of malignancy. Among multiple pulmonary nodules, pulmonary nodules in the right upper lobe (RUL) and left upper lobe had mild fluorodeoxyglucose (FDG) uptake (maximum standardized uptake value [SUVmax] = 3.1 and 2.3, respectively) on F-18 FDG positron emission tomography (PET)/CT (Figure 2). For pathological confirmation, video-assisted thoracoscopic wedge resection of the RUL of the lung was done. Histologically, the tumor consisted of low-grade spindle cells with frequent pseudoinclusions forming storiform and interlacing bundles in a collagen-rich matrix and multifocal psammomatous calcifications (Figure 3). Tumor cells showed positive immunohistochemical (IHC) staining for

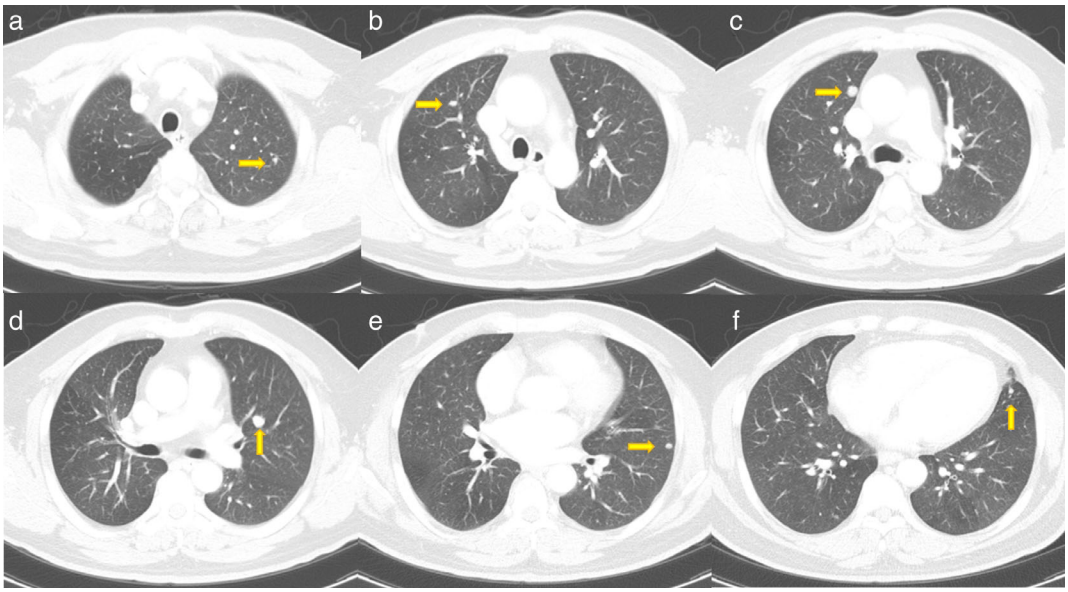

FIGURE 1 Chest CT images showing multiple variable-sized nodules in both lungs (arrows) in lung window-setting views; (a) 5.2 mm on LUL; (b) 5.8 mm on RUL; (c) 9.6 mm on RUL; (d) 12.5 mm on LUL; (e) 6.0 mm on LUL; (f) 6.2 mm on LUL

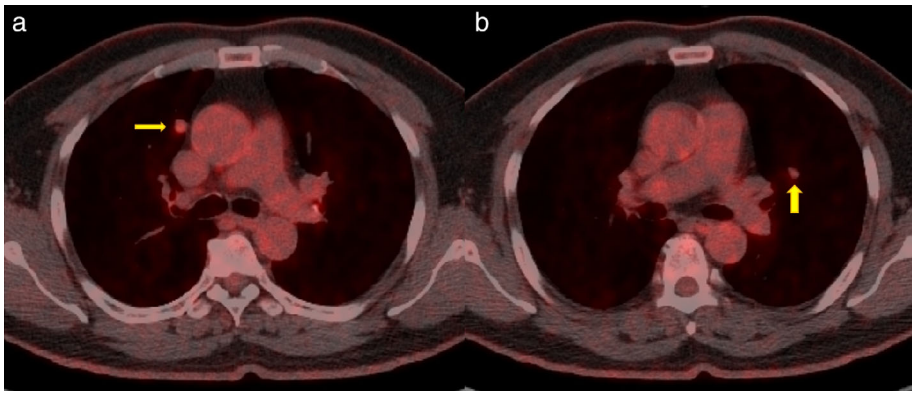

FIGURE 2 Hypermetabolic nodules on PET/CT (arrows) in the right upper lobe; (a) and left upper lobe; (b) Wedge resection of the right upper lobe of the lung, including the nodules, was performed

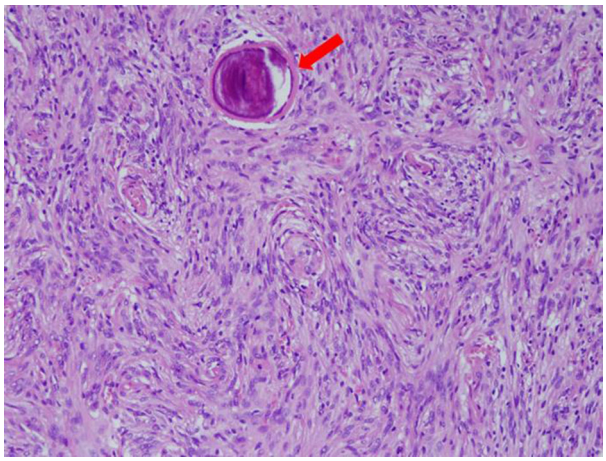

FIGURE 3 Intrapulmonary meningioma (H&E). Whorling spindle cells with pseudoinclusions (red arrow), collagenous matrix, and psammomatous calcification support the diagnosis of meningioma

epithelial membrane antigen (EMA). Brain magnetic resonance imaging revealed no intracranial tumor. He was finally diagnosed with PPM, a benign tumor. The patient has been closely monitored without further interventions, and the nodules have not shown any changes over 24 months.

DISCUSSION

Meningioma originating from the lung is very rare. PPM is usually found incidentally on X-ray images or CT scans, with no typical respiratory symptoms.[4,5] On chest CT, PPM usually presents as a round, well-demarcated, solitary nodule that ranges in size from 0.6 cm to 6 cm.[4,6] However, unlike most previous reported cases of PPM identified as solitary nodules, our case showed multiple lung nodules on chest CT, which led us to suspect them to be metastatic lesions. A previous study described PPM as multiple pulmonary

nodules in 1998.[7] Since then, to the best of our knowledge, multiple nodule cases of PPM have not been reported except for the cases reported by Wang et al. that presented with multiple cystic lesions.[8] In our study, there were several nodules of various sizes in both lungs with a slight enhancement on chest CT mimicking disseminated pulmonary metastases.[9] We therefore attempted to identify the tumor origins from outside the lung by conducting various examinations, including imaging studies and tumor markers before final tissue confirmation. Therefore, as it is difficult to distinguish PPM from lung tumors through radiological findings only, pathological examination for the suspicious lesion is important for the diagnosis of PPM.

In cytology specimens of PPM, characteristic features such as spindle cells in a whorl formation and many psammoma bodies are commonly shown, and specific IHC findings are also useful for excluding incorrect diagnoses.[6,10] Ohashi-Nakatani et al. reported the IHC outcomes of 58 patients with PPM in which Vimentin was 100% positive; EMA and progesterone receptor was approximately 80% and CD34 was approximately 22%. In the present study, the tumor cell showed positive staining for EMA and negative staining for CD34.

A PET/CT scan was performed in our case and FDG uptake was mildly increased as previous reports showing faint FDG avidity.[5] However, in the other two cases of benign PPM, SUVmax was increased up to 12.9 and 10.1, which were suspicious of malignancy.[11,12] Because of the possibility of false-positive PET, detection of FDG uptake on PET/CT scan might not be helpful in determining whether or not PPM is benign.

PPM mostly has a good prognosis without recurrence and metastasis.[13] The main strategy for treatment is surgical resection of the lung and wedge resection or lobectomy for benign PPM is usually performed.[4] In this case, because the patient had several nodules in both lungs, we could not resect all suspicious lesions. The remaining nodules were unchanged on follow-up chest CT for 2 years. However, when considering the report by Satoh et al. which presented the 20-year follow-up findings of remnant PPM lesions exhibiting slow growth with a doubling time of 1393 days, it is imperative to consider the long-term follow-up of several years.[14]

In conclusion, PPM could have a variety of radiological findings. We should therefore consider the possibility of PPM in the differential diagnosis of lung nodules. Furthermore, pathological confirmation is important in the diagnosis of PPM. Because PPM generally has a stable course, careful observation and surgical resection can be options for safe treatment.

CONFLICT OF INTEREST

The authors declare that they have no competing interests to disclose.

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REFERENCES

1. Lamszus K. Meningioma pathology, genetics, and biology. J Neuropathol Exp Neurol. 2004;63:275–86.
2. Rushing EJ, Bouffard JP, McCall S, Olsen C, Mena H, Sandberg GD, et al. Primary extracranial meningiomas: an analysis of 146 cases. Head Neck Pathol. 2009;3:116–30.
3. Kemnitz P, Spormann H, Heinrich P. Meningioma of lung: first report with light and electron microscopic findings. Ultrastruct Pathol. 1982; 3:359–65.
4. Han D, Deng H, Liu Y. Primary pulmonary meningiomas: report of two cases and review of the literature. Pathol Res Pract. 2020;216:153232.
5. Chakrabarti R, Ghuman D. Diagnostic and management considerations in a patient with primary pulmonary meningioma with associated micro-solid nodules. Cureus. 2021;13:e15700.
6. Lin D, Yu Y, Wang H, Fang Y, Yin J, Shen Y, et al. Radiological manifestations, histological features and surgical outcomes of pulmonary meningothelial proliferation: a case series and rethinking. Transl Lung Cancer Res. 2020;9:1159–68.
7. Ueno M, Fujiyama J, Yamazaki I, Uchiyama T, Ishikawa Y, Satoh Y. Cytology of primary pulmonary meningioma. Report of the first multiple case. Acta Cytol. 1998;42:1424–30.
8. Wang X, Li P, Zhou P, Fu Y, Lai Y, Che G. Intrapulmonary metastasis from primary pulmonary meningioma presenting as multiple cystic lesions: a case report. BMC Pulm Med. 2019;19:8.
9. Davis SD. CT evaluation for pulmonary metastases in patients with extrathoracic malignancy. Radiology. 1991;180:1–12.
10. Ohashi-Nakatani K, Shibuki Y, Fujima M, Watanabe R, Yoshida A, Yoshida H, et al. Primary pulmonary meningioma: a rare case report of aspiration cytological features and immunohistochemical assessment. Diagn Cytopathol. 2019;47:330–3.
11. Portes Meirelles GS, Ravizzini G, Moreira AL, Akhurst T. Primary pulmonary meningioma manifesting as a solitary pulmonary nodule with a false-positive PET scan. J Thorac Imaging. 2006;21:225–7.
12. Cura M, Smoak W, Dala R. Pulmonary meningioma: false-positive positron emission tomography for malignant pulmonary nodules. Clin Nucl Med. 2002;27:701–4.
13. Incarbone M, Ceresoli GL, di Tommaso L, et al. Primary pulmonary meningioma: report of a case and review of the literature. Lung Cancer. 2008;62:401–7.
14. Satoh Y, Ishikawa Y. Multiple primary pulmonary meningiomas: 20-year follow-up findings for a first reported case confirming a benign biological nature. Int J Surg Case Rep. 2017;31:58–60.

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