

# **Primary pulmonary meningioma** A case report

Shucheng Huang, MM<sup>a</sup>, Li Chen, MM<sup>a</sup>, Yuping Mao, MM<sup>b</sup>, Hongwei Tong, MM<sup>a,\*</sup>

# Abstract

Rationale: Primary extracranial meningiomas are rare outside the head and neck region.

Patient concerns: A 44-year-old female patient had chest pain for more than 1 year.

**Diagnoses:** Preoperative chest computed tomography (CT) scan revealed a nodule in the right lower lobe, 1.8 cm in diameter. Tumor tissues were examined by immunohistochemistry for vimentin and S-100.

**Interventions:** Histopathologically, the tumor was characterized by whorled nests of spindle-shaped cells accompanied by psammoma bodies. Immunohistochemistry demonstrated tumor cell positivity for vimentin and S-100. This case was diagnosed as a primary pulmonary meningioma. The tumor was removed by a thoracoscopic pulmonary wedge resection.

**Outcomes:** Postoperative cranial and spinal CT scan did not show any intracranial or spinal mass. No recurrence of the tumor was reported at the time of writing up this case report.

Lessons: A primary pulmonary meningioma should be considered in the differential diagnosis workup of pulmonary nodules.

**Abbreviations:** CNS = central nervous system, CT = computed tomography, MRI = magnetic resonance imaging, VATS = video-assisted thoracic surgery.

Keywords: immunohistochemistry, primary pulmonary meningioma, thoracoscopic pulmonary wedge resection

### 1. Introduction

A meningioma is a common primary tumor in the central nervous system (CNS). An ectopic primary meningioma, which accounts for 1% to 2% of all primary meningiomas, is typically seen in head and neck, the cranium, the orbit, the nose, the paranasal sinus, and the oropharynx,<sup>[1,2]</sup> and primary extracranial meningiomas are rare outside the head and neck region. Primary pulmonary meningiomais very rare and so far only ~30 cases have been reported in the English literature since its first report by Kemnitz and Heinrich in 1982.<sup>[3–5]</sup> A primary pulmonary meningioma typically presents as a solitary pulmonary nodule, which is usually benign, and has an excellent prognosis. We report a case of a primary pulmonary meningioma in a 44-year-old female who was successfully managed with thoracoscopic pulmonary wedge resection.

Editor: Levent Dalar.

Medicine (2017) 96:19(e6474)

Received: 7 November 2016 / Received in final form: 14 February 2017 / Accepted: 2 March 2017

http://dx.doi.org/10.1097/MD.00000000006474

# 2. Case report

Approval for the study by the local institutional review board was not required because it was a case report. A 44-year-old female patient was admitted on 7 April 2015 because of chest pain for more than 1 year. The pain was dull, nonradiating, and intermittent and was located in the lower sternum. No cough or hemoptysis was reported. The patient experienced no decreased body weight and fatigue and had no palpitations. The patient also denied a history of tuberculosis, diabetes mellitus, hypertension, or coronary heart disease. Physical examination showed even and unlabored respiration, and no cyanosis in the mouth or lip and no clubbing finger were noticed. There were no palpable enlarged lymph nodes. No deviation of the trachea was noticed. Laboratory findings are shown in Table 1. Both lungs were clear on auscultation with no rales. Chest plain and contrastenhanced computed tomography (CT) scan revealed a nodule in the right lower lobe, 1.8 cm in diameter, with calcifications, mild peripheral lobulation, and mild intensification after contrast enhancement (Fig. 1A and B). No apparent lymph node enlargement was noticed in the hilum and mediastinum. Brain MRI revealed no abnormality.

On 14 April 2015, the patient provided a written informed consent to and underwent thoracoscopic pulmonary wedge resection of the lower right lobe under general anesthesia. Intraoperative exploration showed no pleural adhesion or effusion. A peripheral mass,  $\sim 2 \times 1 \text{ cm}$  in size, was found in the right lower lobe. It was well circumscribed with a distinct border. Intraoperative pathological examination of snap frozen sections revealed a tumor consisting of spindle-shaped cells, which exhibited a fascicular arrangement or concentric whorls. Conventional pathological examination disclosed an ectopic meningioma of the lower right lung, which was  $2.5 \times 1.5 \times 1 \text{ cm}$  in size. Microscopically, the tumor showed whorl formations,

The authors have no conflicts of interest to disclose.

<sup>&</sup>lt;sup>a</sup> Department of Cardiothoracic Surgery, <sup>b</sup> Department of Pathology, Zhejiang Provincial Hospital of Traditional Chinese Medicine, Hangzhou, China.

<sup>&</sup>lt;sup>\*</sup> Correspondence: Hongwei Tong MM, Department of Cardiothoracic Surgery, Zhejiang Provincial Hospital of Traditional Chinese Medicine, Hangzhou, China (e-mail: szxw901@163.com).

Copyright © 2017 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the Creative Commons Attribution-No Derivatives License 4.0, which allows for redistribution, commercial and non-commercial, as long as it is passed along unchanged and in whole, with credit to the author.

Table 1	
Patient demographic data and laboratory findings.	
Age, y	44
Gender	Female
Clinical chemistries	
Glucose, mmol/L	4.28 (3.89–6.11)*
Creatine, mol/L	58.00 (45–84)*
Potassium, mmol/L	3.97 (3.5–5.3)*
Sodium, mmol/L	139.5 (137.0–147.0)*
Albumin, g/L	37.90 (40.0–55.0)*
Radiological findings	
Chest CT scan	Right lower lobe nodule, 1.8 cm in
	diameter, with calcifications, mild peripheral
	lobulation, and mild intensification after
	contrast enhancement
Brain MRI	Unremarkable
Immunohistochemical findings	
Cytokeratin, CK5/6, CK7,	-
TTF-1, P40, CD117, Bcl-2,	
CD68, desmin, calretinin	
P63	+/
P53	+ (5%)
Ki-67	+ (10%)
Vimentin, EMA, CD34, CD31,	+
F8 and SMA (vessel),	
CD163, S-100	

CD = cluster of differentiation, CK = cytokeratin, CT = computed tomography, MRI = magnetic resonance imaging, TTF-1 = thyroid transcription factor-1.

\* Normal reference values.

with cellular atypia and mitotic figures (<3/10 high power fields) (Fig. 2). In addition, psammoma bodies were seen. The tumor extended into the encapsulation. Immunohistochemical findings were as follows: cytokeratin (CK, broad spectrum) (–); CK5/6 (–); CK7 (–);thyroid transcription factor-1 (–); vimentin (+); the epithelial membrane antigen (EMA) (+); P40 (–); P63 (+/–); CD117 (–); CD34, CD31, F8, and smooth muscle actin (vessel +); Bcl-2 (–); CD68 (–); CD163 (+); desmin (–);S-100 (+); P53 (+, 5%); Ki-67 (+, 10%); and calretinin (–). Because postoperative cranial and spinal CT scan did not show any intracranial or spinal mass, this case was diagnosed as a primary pulmonary meningioma. At 6 months of follow-up, the patient was well and showed no evidence of a cranial or a spinal meningioma. No recurrence of the tumor was reported at the time of writing up this case report.

#### 3. Discussion

A primary pulmonary meningioma is extremely rare, but may mimic any other pulmonary tumor presenting as a solitary



Figure 2. A section of the yellowish-white portion of the tumor tissue showing elongated spindle-shaped cells and a delicate fibroconnective tissue arranged in whorls (H&E,  $\times$ 100). Psammoma bodies are also observed.

pulmonary nodule.<sup>[6]</sup> Though a primary pulmonary meningioma may remain asymptomatic and be found incidentally,<sup>[7,8]</sup> atypical chest pain was present for more than 1 year in our case. Kim et al<sup>[4]</sup> have also recently reported a case of a primary pulmonary meningioma in a 61-year-old female with a typical chest pain. A primary pulmonary meningioma is largely benign, grows slowly, and has an excellent prognosis. Satoh and Ishikawa<sup>[9]</sup> followed up a primary pulmonary meningioma patient for 10 years and found no recurrence of the disease following surgical resection. However, a malignant primary pulmonary meningioma has been reported.<sup>[10,11]</sup> Incarbone et al<sup>[12]</sup> reviewed 37 cases of primary pulmonary meningiomas reported in the literature. Among 25 histologically confirmed primary pulmonary meningioma patients with radiological data of the central nervous system and histological assessment, 23 (92%) cases had benign and 2 (8%) cases had a malignant primary pulmonary meningioma. Though our case appears to be benign, the tumor has some features usually associated with malignant cancer such as cellular atypia, prominent nuclei, occasional mitotic figures, and penetration through the encapsulation.

Like most reported cases of a primary pulmonary meningioma, the tumor in our patient was also largely well circumscribed. Such tumors are amenable to surgical resection and seldom recur postoperatively. Incarbone et al<sup>[12]</sup> suggested that when feasible, pulmonary wedge resection by video-assisted thoracic surgery should be performed. Our patient underwent thoracoscopic



Figure 1. Plain (A) and contrast CT scan (B) of the lungs of a 44-year old female patient diagnosed with a primary pulmonary meningioma. CT = computed tomography.

pulmonary wedge resection of the right lower lobe and had no recurrence so far.

Although rare, a primary pulmonary meningioma should be considered in the differential diagnosis of pulmonary nodules. Macroscopically, a primary pulmonary meningioma typically manifests as round, well-defined pulmonary nodules. Immunohistochemistry demonstrated tumor cell positivity for vimentin and EMA, as is the characteristic of a meningioma. Microscopically, the tumor cells are arranged in sheets, whorls, or onion peel-like formations. Diagnosis of a primary pulmonary meningioma relies on the presence of the tumor in the lungs, exclusion by CT or MRI of a meningioma in the CNS, and pathological and immunohistochemical features consistent with a primary pulmonary meningioma. Therefore, diagnosis of the disease requires pathological, immunochemical, and radiological studies.

In conclusion, a primary pulmonary meningioma is a rare disease and typically benign as illustrated in the present case. It should be considered in the differential diagnosis workup of pulmonary nodules.

#### References

 Muzumdar DP, Vengsarkar US, Bhatjiwale MG, et al. Diffuse calvarial meningioma: a case report. J Postgrad Med 2001;47:116–8.

- [2] Wu ZB, Yang GH. People's Medical Publishing House Chinese Surgical Pathology [Book in Chinese] 2002:2544-5.
- [3] Kemnitz PSH, Heinrich P. Meningiom a of lung: first report with light and electronmicroscopic findings. Ultrastruct Pathol 1982;3:359–65.
- [4] Kim YY, Hong YK, Kie JH, et al. Primary pulmonary meningioma: an unusual cause of a nodule with strong and homogeneous enhancement. Clin Imaging 2016;40:170–3.
- [5] Masago K, Hosada W, Sasaki E, et al. Is primary pulmonary meningioma a giant form of a meningothelial-like nodule? A case report and review of the literature. Case Rep Oncol 2012;5:471–8.
- [6] Meirelles GS, Ravizzini G, Moreira AL, et al. Primary pulmonary meningioma manifesting as a solitary pulmonary nodule with a falsepositive PET scan. J Thorac Imaging 2006;21:225–7.
- [7] Kaleem Z, Fitzpatrick MM, Ritter JH. Primary pulmonary meningioma. Report of a case and review of the literature. Arch Pathol Lab Med 1997;121:631–6.
- [8] Chiarelli M, De Simone M, Gerosa M, et al. An incidental pulmonary meningioma revealing an intracranial meningioma: primary or secondary lesion? Ann Thorac Surg 2015;99:e83–4.
- [9] Satoh Y, Ishikawa Y. Primary pulmonary meningioma: ten-year followup findings for a multiple case, implying a benign biological nature. J Thorac Cardiovasc Surg 2010;139:e39–40.
- [10] Prayson RA, Farver CF. Primary pulmonary malignant meningioma. Am J Surg Pathol 1999;23:722–6.
- [11] van der Meij JJ, Boomars KA, van den Bosch JM, et al. Primary pulmonary malignant meningioma. Ann Thorac Surg 2005;80:1523–5.
- [12] 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.