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Primary pulmonary meningioma presenting as a pulmonary ground glass nodule: a case report and review of the literature

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Abstract

Background Primary pulmonary meningioma (PPM) is an extremely rare entity. PPM manifested with GGN are a very rare occurrence in clinical practice.

Case presentation In this study, we report a case of PPM with atypical computed tomography (CT) features. A 59 year old Han Chinese female came to our hospital for treatment and reported that her physical examination revealed GGN in the right lung for over 3 months. The histologic result revealed a PPM. The patient underwent a thoracoscopic lung wedge resection of the right upper lobe for a GGN. After 1 years of follow-up, the patient is still alive without evidence of metastasis or recurrence.

Conclusions PPM could have a variety of radiological findings. As there are no specific radiologic features for the diagnosis of PPM, complete resection of the lesion is required for both diagnosis and treatment. It is necessary to note the imaging features of PPM, presenting as a GGN; this rare tumor should be considered in differential diagnoses.

Keywords Primary pulmonary meningioma, Pulmonary ground glass nodule, Thoracoscopic pulmonary wedge resection

Introduction

A meningioma is a common primary tumor in the central nervous system (CNS). An ectopic primary meningioma, which accounts for 1–2% of all primary meningiomas, is rare: occurs in several locations, such as the head-and-neck region, skin, bone, peripheral nerves, retroperitoneum and lung [1, 2].Primary pulmonary meningioma (PPM) is a very rare and mostly benign disease. Few previous studies have reported the clinical features of PPM, which has led to the low diagnostic rate of early PPM. Rates of misdiagnosis and missed diagnosis are relatively high in clinical practice. PPM manifested with GGN is a very rare occurrence in clinical practice. Recently, a patient with PPM and GGN was diagnosed and treated at our medical center. This paper describes this rare form of PPM through a case report and literature review.

Case presentation

A 59 year old Han Chinese female came to our hospital for treatment and reported that her physical examination revealed GGN in the right lung for over 3 months. Chest CT was performed at our hospital, revealing a 4.0–5.0-mm GGN in the right upper lobe (Fig. 1). Due to the small size and low density of the GGN, we first suspected it to be a pneumonia nodule. After anti-inflammatory treatment for half a month, the chest CT reexamination showed that the GGN still existed. The patient had no

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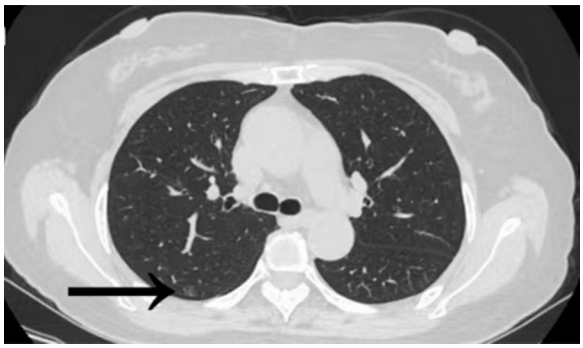

Fig. 1 Chest CT scan showing a 4–5 mm GGN in the right upper lobe (arrowheads)

history of chronic disease, malignant tumor, smoking, tuberculosis, or cancer in the family. The results of the laboratory tests were within normal limits. The central nervous system (CNS) was normal on magnetic resonance imaging (MRI). Also, no abnormality in the central nervous system has been recognized.

A thoracoscopic wedge resection of the GGN was performed with an intraoperative frozen section, revealing a spindle-cell tumor. Immunohistochemically the tumor cells showed staining positive for epithelial membrane antigen (EMA) and vimentin, whereas they were negative for keratin, S-100 protein and neuron-specific enolase (Fig. 2). Medium power photomicrograph of the tumor showing spindle-shaped cells with poorly defined cell borders arranged in whorls (Fig. 3).

Finally, a histological diagnosis of PPM without characteristics of malignancy was made according to the

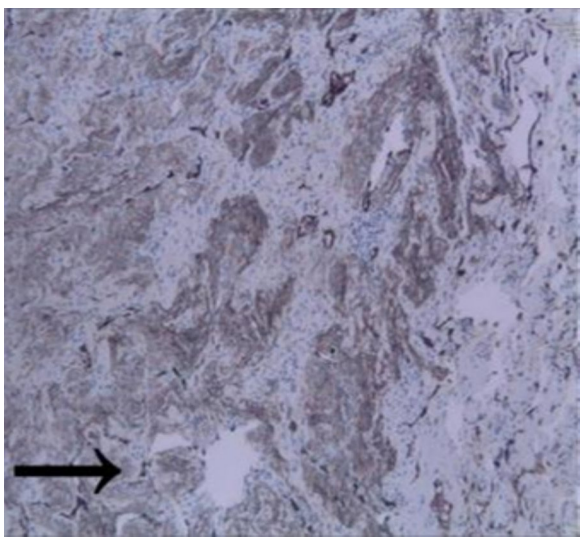

Fig. 2 Immunohistochemically (IHC) the tumor cells shows staining positive for epithelial membrane antigen (EMA), ×200 (arrowheads)

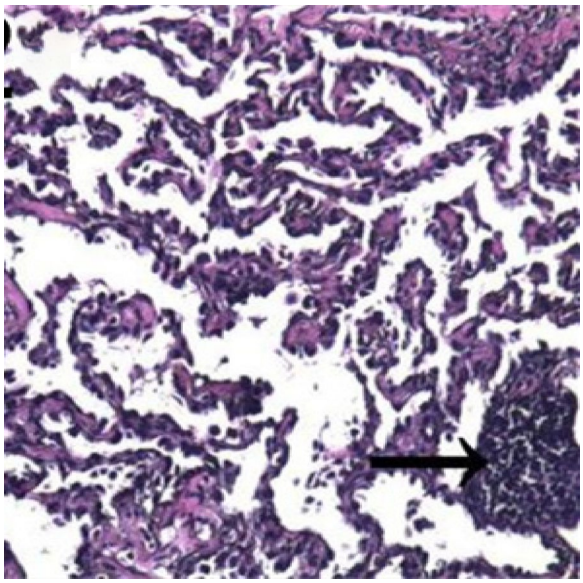

Fig. 3 Medium power photomicrograph of the tumor shows spindle-shaped cells with poorly defined cell borders arranged in whorls, Hematoxylin & eosin, ×100 (arrowheads)

above-described morphological and immunohistochemical features. After 1 years of follow-up, the patient is still alive without evidence of metastasis or recurrence. The patient agreed to authorize us to share the figures and the experiences during his treatment procedure in our department. Informed consent was obtained.

Discussion

Reports of meningiomas that primarily originate outside the CNS are very rare. Thus far these meningiomas have been detected in following sites: intra-orbital, scalp and subcutaneous tissue, skull, nasal sinus, intraparenchymal, epidural, parotid, thorax, adrenal, and fingers [3]. Meningiomas that primarily originate from the lung are more rare. There are several controversial hypotheses regarding the pathogenesis of PPM. For instance, some authors have proposed that the tumor develops from minute pulmonary meningothelial-like nodules [4], whereas others suggest that the tumor may arise from pluripotent subpleural mesenchyme [5]. The true etiology of this tumor is still uncertain [6]. A total of 68 patients diagnosed with PPM were reported in the English literature from 1982 to 2021 [7, 8].

PPM is a rare disease that is usually asymptomatic and primarily occurs in 40–60 year old patients. Nevertheless, patients with larger tumors may experience chest pain and other symptoms [9]. In our case, the patient did not have any clinical symptoms, and GGN was found during physical examination. In the vast majority of patients, pulmonary masses and nodules are detected during

physical examination. X-rays and CT scans usually reveal round or oval solitary nodules or mass shadows of different size, uniform density, smooth boundaries, and a clear outline [10]. Previous reports of PPM with contrast-enhanced CT images showed various enhancement patterns, such as heterogeneous or nodular enhancement [11], poor enhancement [12], or homogeneous enhancement [7].

In our case, it is very rare for PPM patients to have GGN on chest CT. Because of its small size, low density and blurry edge, we considered it as a pneumonia node in the early diagnosis, but not excluding the possibility of atypical adenomatous hyperplasia, adenocarcinoma in situ, and minimally invasive adenocarcinoma. We reviewed chest CT half a month after anti-inflammatory treatment, and GGN was still present. The patient's lung nodule was a sub centimeter (4–5 mm) nodule. As per the guidelines and surveillance protocol no surgical intervention is indicated. However, the patient was in a serious state of anxiety and depression, and we considered that the patient had "pulmonary nodule syndrome". In order to alleviate the psychological state of anxiety and depression, we obtained the consent of the patient's family, so we performed surgical resection, and the PPM was confirmed after surgery. Distinguishing PPM from other lung tumors may be difficult, as the most common manifestation is isolated GGN. Therefore, the early diagnosis of such GGN is difficult. Pathological identification is necessary for a final diagnosis of PPM. In many cases, an enhanced CT scan shows a mass with different degrees of non-uniform enhancement, distinguished from uniform and apparent enhancement in an intracranial meningioma [10]. Vimetin and EMA are simultaneously expressed in the majority of patients. CD34 foci are positive in individual cases, while keratin, CK, and S-100 proteins are negative [13].

PPM mostly has a good prognosis without recurrence and metastasis [14]. The main strategy for treatment is surgical resection of the lung and wedge resection or lobectomy for benign PPM is usually performed. 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 [15]. Malignant PPMs with aggressive growth and distant metastases are extremely rare. However, there are also reports of malignant PPMs. Prayson et al. reported a very aggressive case of PPM. Ipsilateral lobe metastases were detected in a patient six months after surgery [16].

In conclusion, PPM could have a variety of radiological findings. As there are no specific radiologic features for the diagnosis of PPM, complete resection of the lesion is required for both diagnosis and treatment. It is necessary to note the imaging features of PPM, presenting as a GGN; this rare tumor should be considered in differential diagnoses.

Abbreviations

PPM	Primary pulmonary meningioma
GGN	Ground glass nodule
CT	Computed tomography
CNS	Central nervous system
EMA	Epithelial membrane antigen
MRI	Magnetic resonance imaging

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None.

Author contributions
ZSL and LXQ analyzed and interpreted the patient data regarding PPM.XMZ, DJG and ZSL performed for the surgery. ZSL and DJG were major contributors in writing the manuscript.

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Availability of data and materials
All patient records, operation notes, and radiographic information are available in the form of hard copies. Scanned documents can be provided upon request from the journal.

Code availability
Not applicable.

Declarations

Ethics approval and consent to participate
Not applicable. Written informed consent was obtained from the patient, and a scanned copy of the consent form can be provided upon request from the journal.

Consent for publication
Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests
The authors declare that they have no competing interests. All authors unanimously agreed to provide consent to publish this paper in the journal.

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