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Primary pulmonary meningioma: an unusual cause of a nodule with strong and homogeneous enhancement☆

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ABSTRACT

We report a case of a 61-year-old female with atypical chest pain. The chest CT scan revealed a well-circumscribed large intrapulmonary nodule that showed vigorous and homogeneous contrast enhancement. The nodule was diagnosed as a meningioma after surgery. Metastatic meningioma was excluded by brain and spine MRI scans. Primary pulmonary meningioma usually appears as a solitary well-defined round or lobulated nodule with variable enhancement on CT; this case is unique because of the intense and homogeneous enhancement. Although rare, primary pulmonary meningioma should be considered in the differential diagnosis of a well-defined pulmonary nodule with dense and homogeneous enhancement.

1. Introduction

Ectopic meningiomas occur rarely in several locations, including the head and neck region, skin, peripheral nerves, retroperitoneum, pelvis, and thorax [1–6]. There are only 44 case reports of primary pulmonary meningioma in the English literature based on a PubMed search using the following terms: primary[All Fields] AND ("lung"[MeSH Terms] OR "lung"[All Fields] OR "pulmonary"[All Fields]) AND ("meningioma"[MeSH Terms] OR "meningioma"[All Fields]). The first reported case was in 1982 [7]. Among the listed cases, only five were malignant or atypical meningiomas [8–12]. Primary pulmonary meningiomas are most likely to be benign.

Based on a systematic review of reported imaging findings, primary pulmonary meningioma usually appears as a solitary pulmonary nodule on chest radiographs and a sharply marginated round or lobulated nodule without calcification on computed tomography (CT) scans. In six previous case reports that used contrast-enhanced CT images, the tumors showed variable enhancement patterns. Here, we present a unique case of an avidly and homogeneously enhancing primary pulmonary meningioma.

2. Case report

The institutional review board (IRB) of our hospital approved this case study and waived the requirement for the subject's informed consent. A 61-year-old woman visited our hospital complaining of atypical chest pain of 2 years' duration that had worsened a month earlier. She had a history of hypertension and diabetes and did not smoke. The results of electrocardiography and a treadmill test were negative which excluded coronary artery diseases. Chest radiographs revealed a well-defined large solitary pulmonary nodule in the right upper lobe (Fig. 1). A chest CT scan with contrast enhancement was performed to better characterize the lesion. A 2.5-cm diameter nodule with a well-circumscribed margin was observed in the anterior segment of the right upper lobe. The nodule was confined to the lung parenchyma. After contrast enhancement, the lesion showed marked homogeneous enhancement of more than 100 Hounsfield units (HUs), from a precontrast attenuation of 40 HU to a postcontrast of 180 HU, suggesting a highly hypervascular tumor (Fig. 2). The results of laboratory tests were within normal limits.

A right upper lobectomy and mediastinal lymph node dissection were performed for pathologic confirmation and possible chest pain alleviation. The operation was terminated after frozen section biopsy revealed that the surgical specimen was benign. The lesion was an oval to round, pinkish-tan intrapulmonary solid mass (Fig. 3A). The pathologic examination revealed meningothelioid cellular nests surrounded by thin collagenous septa consistent with a meningioma (Fig. 3B). There was no tumor involvement in the dissected lymph nodes.

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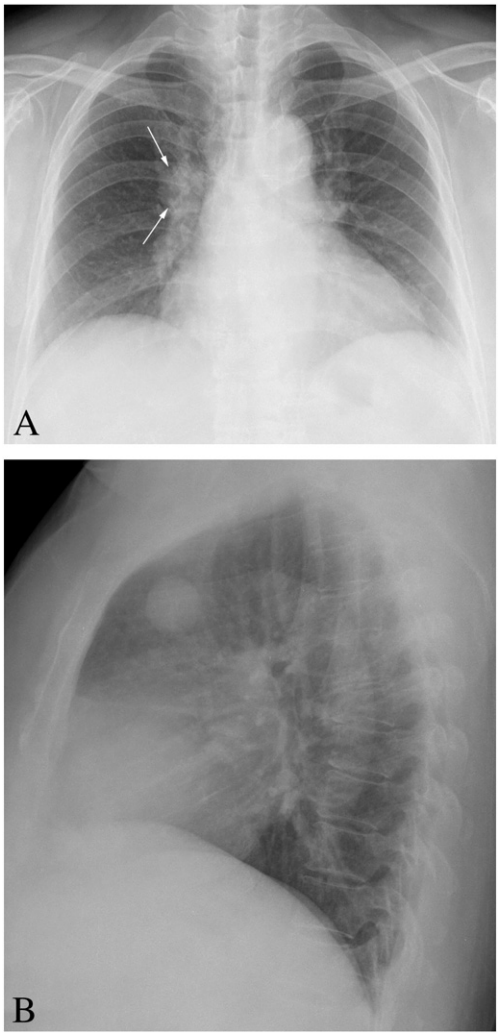

Fig. 1. Chest radiographs of (A) posteroanterior (PA) and (B) lateral view. There is a well-demarcated large nodule in the right upper lobe, coalescent with a hilar opacity on the PA radiograph (arrows).

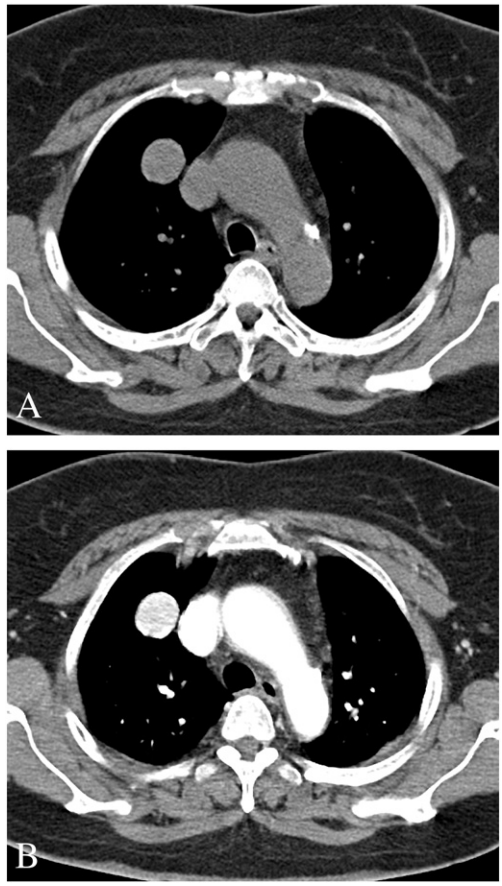

Fig. 2. Chest CT images of (A) unenhanced and (B) enhanced scan. A 2.5-cm diameter well-circumscribed nodule in the anterior segment of the right upper lobe shows strong and homogeneous enhancement.

To exclude metastatic meningioma, brain and whole spine magnetic resonance imaging (MRI) scans were performed after the operation. There was no evidence of intracranial or spinal meningioma. The patient was therefore diagnosed with an isolated primary pulmonary meningioma. Her symptoms improved after the surgery, and she is doing well without tumor recurrence for 7 years.

3. Discussion

Primary pulmonary meningioma is a rare disease, with only 44 cases reported in the English language. Intrapulmonary metastasis of meningioma is also rare. Less than 1% of intracranial meningioma cases metastasize, most commonly to the lung [13]. The fact that some previous reports of pulmonary meningioma did not refer to imaging studies to exclude metastasis [14–17] makes primary pulmonary meningioma even rarer. The pathogenesis of primary pulmonary meningioma remains controversial. One hypothesis is that the tumor develops from minute pulmonary meningothelial-like nodules (MPMNs) incidentally found in about 1% of autopsies [18]. However, a recent genotypic comparison recognized molecular differences between MPMNs and meningiomas, disputing the hypothesis [19]. Pluripotential subpleural mesenchyme is another possible origin [20].

Meningioma is the most common primary intracranial tumor [21]. The typical imaging findings of intracranial meningioma are well established. Most tumors have a sharply circumscribed morphology and a homogeneous and avid enhancing pattern on CT or MRI in typical extra-axial locations such as the cerebral convexity, parasagittal region, or skull base [22,23]. Interestingly, among the six prior reports of primary pulmonary meningioma with contrast-enhanced CT images, two showed a heterogeneous or nodular enhancement pattern [24,25] and two cases were poorly enhanced [26,27]. It is significant that this primary intrapulmonary meningioma case and two other cases with homogeneous enhancement [28,29] had the same enhancement pattern as that of a typical primary intracranial tumor.

Our case is unique among primary pulmonary meningiomas because of its strong and homogeneous enhancement, which emphasizes that this tumor should be included in the differential diagnosis of well-defined, strongly enhancing intrapulmonary nodules. Other differential diagnoses include typical or atypical carcinoid tumors, Castleman's disease, sclerosing hemangioma, bacillary angiomatosis, pulmonary arteriovenous fistula, and pulmonary varix. Typical or atypical carcinoid tumors, indolent low-grade neuroendocrine malignancies, can be found in the periphery of the lung [30]. This is more common for atypical carcinoids, since typical carcinoids have a predilection for a central location. These tumors usually show a well-circumscribed, lobulated margin and significant enhancement and are related to the airways. The localized form of Castleman's disease or angiofollicular lymph node hyperplasia rarely originates from an intrapulmonary lymph node and appears as a well-marginated nodule that enhances strongly in the centripetal direction where enlarged feeding vessels may be found [31]. Sclerosing hemangioma, a rare benign or low-grade tumor that generally has three or more histologic components, appears as a well-demarcated nodule. It enhances markedly and rapidly when it has a hemangiomatous or papillary histologic component [32].