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CASE REPORT

Primary pulmonary meningioma Report of a case and review of the literature

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Summary Primary pulmonary meningioma (PPM) is a rare disease and usually presents as a solitary pulmonary nodule (SPN). These lesions are mostly benign, but malignant PPMs have been reported, and primary lung cancer or metastasis may be suspected on imaging.

We report one case of benign PPM, with a review of 37 cases published in the literature. Diagnostic work-up included radiological chest study and in 3 cases positron emission tomography (PET) showing increased uptake, highly suspicious for malignancy. After exclusion of 13 cases lacking radiological studies of the central nervous system (CNS), 25 patients with radiological data and histological assessment confirming PPM were considered in the analysis. All patients underwent surgical resection except for 1 case with diagnosis at autopsy.

Histological assessment revealed benign PPMs in 23 cases (including all 3 cases with positive PET) and malignant PPMs in 2 cases. No recurrence was observed in long-term follow-up of patients with benign PPMs, but the two malignant PPMs relapsed.

PPM is an uncommon SPN, so that it can be misdiagnosed and its management unsuited. Indeed, 8 patients (32%) were overtreated with major thoracic surgical resection or with chemotherapy.

When feasible, pulmonary wedge resection by video-assisted thoracic surgery (VATS) including intra-operative histological examination is the most suitable approach to determine the diagnosis and the volume of pulmonary resection.

1. Introduction

Primary extracranial and extraspinal meningiomas are rare tumors and usually occur in the head and neck region or less frequently in the skin and peripheral nerves [1,2]. Primary pulmonary meningiomas (PPMs) are even more rare,

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and since the first description in 1982 by Kemnitz et al. [3] only 36 cases have been reported in the medical literature.

The etiology of these tumors is still uncertain and different histogenetic mechanisms have been proposed. Most PPMs are benign neoplasms, but two malignant cases have been described [4,5]. PPM usually appears as a solitary pulmonary nodule (SPN) that is detected incidentally by chest radiograph or computed tomography (CT).

The radiological evaluation of an indeterminate SPN can be a challenge and indeed, despite the use of diagnostic modalities including CT, positron emission tomography (PET) and needle aspiration biopsy, many benign PPMs were misdiagnosed and overtreated with major pulmonary resection or chemotherapy.

2. Patients and methods

2.1. Case report

A 24-year-old man presented with hemoptysis, after which a SPN on the right upper lobe of the lung was discovered on chest radiograph. Contrast-enhanced CT of the chest and abdomen revealed a 2.4 cm intrapulmonary lesion next to the innominate vein, the appearance of which was suspicious for malignancy [Fig. 1A]. On 18-fluoro-2-deoxy-D-glucose (FDG) PET imaging, the nodule standardized uptake value (SUV) was 10.14, typical for a malignant lesion [Fig. 1B]. No other lesions were detected on CT or FDG-PET. The patient was admitted to our hospital where magnetic resonance angiography showed a cleavage plane between the SPN and the innominate vein [Fig. 1C]. Given the CT and PET features of the nodule, as well as the nodule's location, we decided to perform a minimally-invasive surgical approach. A wedge resection of the right upper lobe was performed using video-assisted thoracic surgery (VATS). An intra-operative frozen histological section excluded the malignancy of the lesion. The whole procedure was completed in 40 min with no complications, and the patient was discharged 2 days later.

Gross examination revealed a 2.4 cm, well-circumscribed, yellowish-red, partially-cystic lesion. Microscopic examination disclosed a tumor compressing the adjacent lung parenchyma as well as several intra-lesional hemorrhagic foci [Fig. 2A]. At higher magnification the lesion was composed of nests of polygonal to spindle-shaped cells arranged in fascicles or whorls. The tumor cells had abundant eosinophilic or clear cytoplasm and round-to-oval nuclei with delicate chromatin distribution and inconspicuous nucleoli [Fig. 2B]. Mitotic figures were absent. On immunohistochemistry, the neoplastic cells stained positive for EMA (Novocastra, GP1.4, 1:400) [Fig. 2C], vimentin (Dako, V9, 1:50) [Fig. 2D] and CD10 (Novocastra, 56C6, 1:100). These morphological and immunohistochemical features were suggestive of a meningioma.

As the central nervous system (CNS) was normal on post-operative magnetic resonance imaging (MRI), the diagnosis of PPM was made. The patient is disease-free after 42 months of follow-up.

2.2. Review of published cases

Thirty-seven patients with a diagnosis of PPM were reported in the literature from 1982 to 2006 [3—28]. All patients reported had histological assessment confirming pulmonary meningioma. Twenty-five patients, including the case we reported above, had radiologic evaluation of the CNS negative for meningioma and were included in the analysis

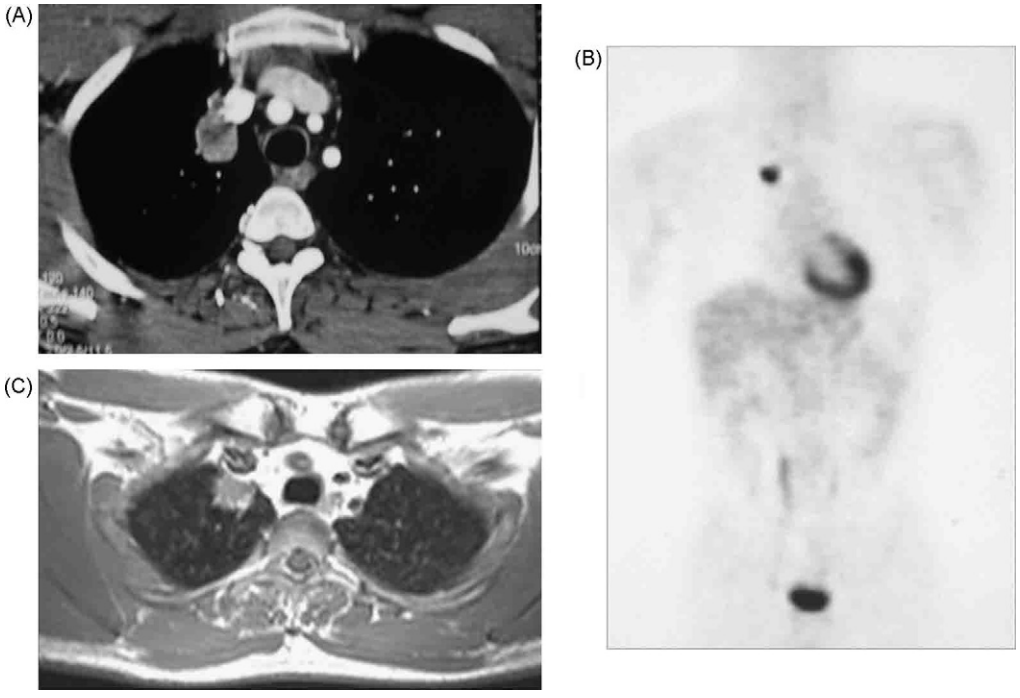

Fig. 1 (A) Suspicious nodular enhancement on chest CT. B) Positive uptake by the nodule on FDG-PET, suggesting malignancy. C) Cleavage plane between the nodule and the innominate vein on magnetic resonance angiography.