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Multiple primary pulmonary meningiomas: 20-year follow-up findings for a first reported case confirming a benign biological nature

Yukitoshi Satoh[a,*], Yuichi Ishikawa[b]

[a] Department of Thoracic Surgery, Kitasato University School of Medicine, 1-15-1 Kitasato, Minami-ku, Sagamihara, Kanagawa 252-0374, Japan
[b] Department of Pathology, Cancer Institute, Japanese Foundation for Cancer Research, 3-8-31 Ariake, Koto-ku, Tokyo 135-8550, Japan

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ABSTRACT

INTRODUCTION: Primary pulmonary meningiomas (PPMs) are very rare neoplasms, and we reported the first multiple case of PPMs in 1998. Usually, PPMs are slow-growing tumors with a good prognosis. Herein, we reported a rare multiple case of PPMs with an extremely long clinical course.
PRESENTATION OF THE CASE: An 84-year-old Japanese woman with a history of multiple PPMs histologically confirmed as having a right-sided slowly growing lung nodule for 20 years. In 2010, we also reported the additional clinical course in the initial case 10 years after surgery implying a benign biological nature. Subsequent to that report, new lesions appeared in the bilateral lung fields with quite a slow growth rate. She was asymptomatic, and no tumor was observed in the cranial cavity during the course of her disease.
DISCUSSION AND CONCLUSION: This case illustrates rare multiple PPMs and highlights their biological behavior with very slow progression from a most likely benign tumor over a 20-year period.

1. Introduction

In 1998, we reported the first multiple case of primary pulmonary meningiomas (PPMs), extremely rare lung neoplasms [1]. Then in 2010, we also reported the additional clinical course in the initial case 10 years after surgery implying a benign biological nature [2]. There have been fewer than 50 cases of PPMs reported in the literature to date [3,4]. Although the lesions are widely known most likely to be benign, grow slowly, and have an excellent prognosis, the etiology is still uncertain; and hence, several mechanisms have been proposed [1,4–8]. Here, we present an additional extremely long course of the initial multiple case of PPM, 20 years after surgery, with further examination by imaging modalities in a specialized institution and then confirm particularly its being benign in nature. The work has been reported in line with the SCARE guideline [9].

2. Case report

An 84-year-old Japanese woman with a history of multiple PPMs, who had undergone surgery for those lesions at the age of 64, was followed up with a chest computed tomography (CT) scan and magnetic resonance imaging examinations. Ten years after the initial surgery, the patient was referred to the Cancer Institute Hospital of the Japanese Foundation for Cancer Research, for a percutaneous biopsy of a new asymptomatic nodule that appeared in a chest CT scan in the right lower lung field, histologically a benign PPM, that showed the same morphologic features as those of the previously reported lesions: transitional meningioma Grade I by the WHO grading system in 2007 (Fig. 1) [1,2]. Immunohistochemical examination demonstrated consistent expression of the epithelial membrane antigen and vimentin in tumor cells. Conversely, results of analyses for the S-100 protein, AE1/AE3, CAM 5.2, muscle-specific actin (HHF-35), smooth muscle actin, synaptophysin, neuron-specific antigen, and desmin were all negative. Furthermore, the tumor cells were focally positive for CD68 by KP-1 and progesterone receptor [2]. Her medical history included hypertension since the first surgery. She was asymptomatic and had no intracranial tumor during all the course of the disease. Based on the morphologic features and the benign clinical course, with sufficient informed consent, the patient is being followed up with periodic CT scans once a year [2]. Although the right lung nodule has grown, the growth is very slow, with a doubling time of 1393 days, the patient is otherwise asymptomatic (Fig. 2). Subsequent to the previous report in 2010 [2], new lesions appeared in the bilateral lung fields with quite a slow growth rate (Fig. 3). These lesions were not histologically evaluated because the patient rejected further invasive and morphologic diagnostic examinations.

Informed consent was obtained from this patient in the report.

* Corresponding author.
E-mail addresses: ysatoh@med.kitasato-u.ac.jp (Y. Satoh), ishikawa@jfcr.or.jp (Y. Ishikawa).

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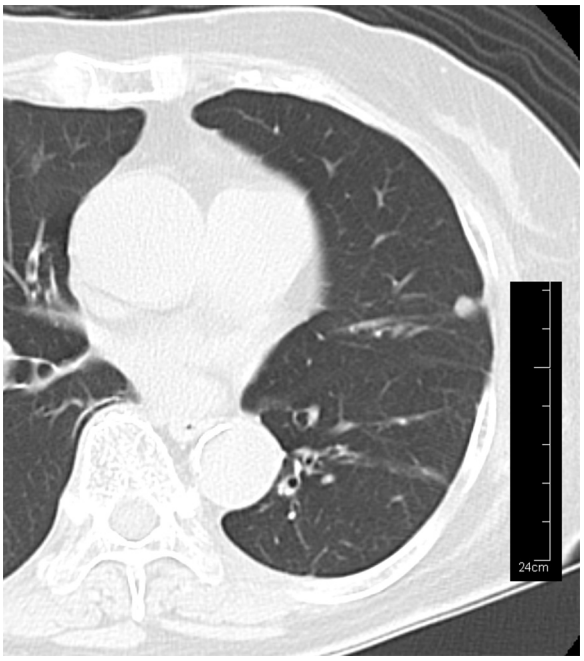

Fig. 1. Microscopically, the right lung nodule consists of tumor cells demonstrating a mixture of meningotheliomatous and fibrous elements (hematoxylin and eosin stain). Note the spindle-shaped cells had ovoid or elongated nuclei and were arranged in palisades intermingled with numerous whorled nests.

3. Discussion

We have successfully followed up the first multiple case of PPMs for more than 20 years after the initial surgery, and, to our knowledge, this is the first case of multiple PPMs with long-term follow-ups. During that period, the possibility of metastasis from an intracranial or intraspinal primary lesion was completely excluded by imaging modalities including a CT scan and an MRI. In this patient, another lesion developed in the right lung that was histologically confirmed to be a meningioma, but the growth rate was extremely slow [2]. After the second report of the case [2], other multiple new lesions had appeared in the bilateral lung fields. These new lesions were determined to be a meningioma based on the clinical course and CT images instead of a biopsy.

A definitive etiology of PPM remains unclear, and at least four different theories have been proposed considering the intrathoracic differentiation of meningocytes or arachnoid cells, the ectopic proliferation of arachnoid cells, tumor development from embryonic stem cells in the embryonic rest theory, or direct/indirect extension of primary intracranial meningiomas [1,7,8]. In the present case, because multiple lesions appeared in the bilateral lungs during the clinical course, the possibility that multiple pulmonary meningothelial-like nodules grew synchronously and metachronously was highly regarded [1,2]. This hypothesis, however, was considered unlikely by other authors, because there was a great discrepancy between the incidence of meningothelial-like nodules and pulmonary meningiomas [6].

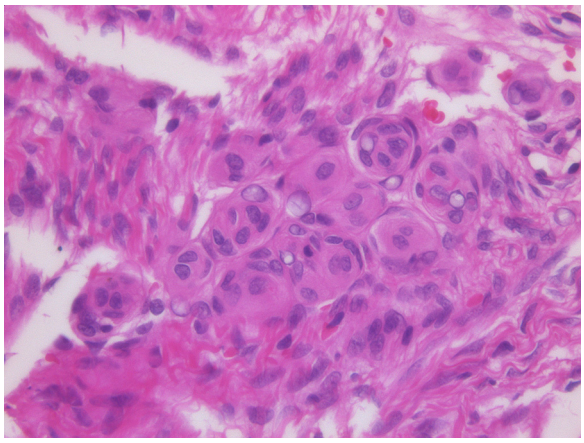

Fig. 3. Chest CT scan showing a 6-mm nodule in the left upper lung field.

These tumors usually grow very slowly, and patients are almost always asymptomatic. Even though the majority of PPM cases are of benign tumors, the treatment usually consists of complete surgical resection. But the recent report described a rare autopsy example in which the tumors had recurred in the lung field, regional lymph nodes, and liver, one even after a 40-year history [6]. Therefore, this case provided unique data on the natural history of apparently benign neoplasms or other long-standing diseases [6].

4. Conclusion

In the present case, even though the multiple new lesions demonstrated continued growth on imaging, a benign native neoplasm was definitely indicated based on the lengthy clinical course. Therefore, we considered that multiple PPMs are most effectively diagnosed by surgical resection then followed, particularly in patients in whom the neoplasms prove to be benign by morphology and extremely long and asymptomatic clinical course.

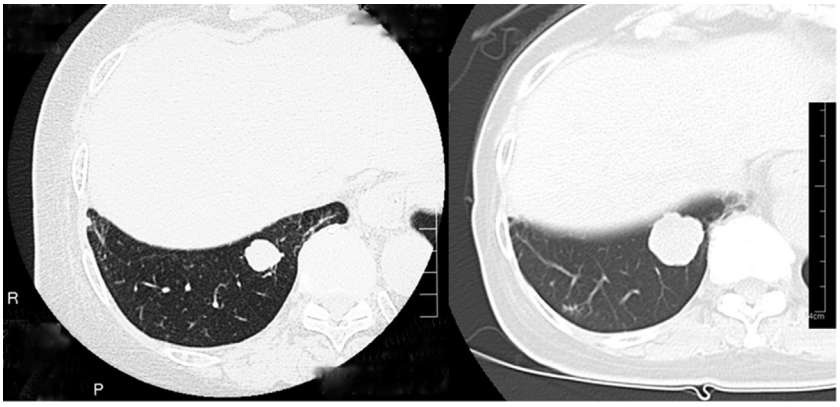

Fig. 2. Chest CT scan showing a well-demarcated nodule in the right lower lung field. The nodule has grown very slowly (left, an 18-mm nodule in 2007; right a 27.5-mm nodule in 2015).

Conflicts of interest statement

The authors have no conflicts of interest Ethic approval not required for the publication of this manuscript.

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Ethics approval

Ethic approval not required.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contribution

Yukitoshi Satoh – Author.
Yuichi Ishikawa – Co-author, revision.

Guarantor

Not applicable.

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