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Case Report

CASE REPORT

Malignant primary pulmonary meningioma with bone metastasis

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Abstract

Primary pulmonary meningioma is extremely rare and only <45 cases have been reported since its first report by Kemnitz and Heinrich (Meningioma of lung: first report with light and electronmicroscopic findings. *Ultrastruct. Pathol.* 1982;3:359–65). Among these cases, only five cases were malignant or atypical. A 67-year-old female with primary pulmonary meningioma underwent thoracoscopic pulmonary wedge resection of the left lower lobe a year before. She had been aware of left thigh pain for 9 months, then she was referred to our division. Positron emission tomography-computed tomography suspected multiple bone metastasis including frontal bone, mandible, left scapula, rib, vertebra, pubis, left femur and sternum. We performed a needle biopsy of the sternum. Histopathological diagnosis was metastasis of primary pulmonary meningioma, World Health Organization grade III. We treated her with denosumab and radiation therapy targeting the left femur. Two months after the treatment, the pain had decreased and she could walk with no cane. No case of malignant extracranial meningioma with bone metastasis have been reported.

INTRODUCTION

A meningioma is a common primary tumor in the central nervous system (CNS). An ectopic primary meningioma (extracranial meningiomas), 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]. Some previous studies reported that extracranial meningioma might derive from misplaced or migrate arachnoid cells, while other previous studies reported that the tumor may originate from perineurial cells or pluripotent mesenchymal cells [2]. The true etiology of this tumor is still uncertain [3]. Primary pulmonary meningioma is extremely rare and so far only <45 cases have been reported in the English literature since its first report by Kemnitz and Heinrich [1, 4–6]. Among these cases, only five cases were malignant or atypical [1, 2]. As far as we investigated, no case of malignant or atypical primary pulmonary meningioma with bone metastasis has been reported.

CASE REPORT

A 67-year-old female with primary pulmonary meningioma underwent thoracoscopic pulmonary wedge resection of the left lower lobe a year before in our hospital. She had been aware of left thigh pain for the last 9 months, then she was referred to our division. X-ray showed osteolytic lesion in the left femur (Fig. 1). Positron emission tomography-computed tomography (PET-CT) suspected multiple bone metastasis including frontal bone, mandible, left scapula, rib, vertebra, pubis, left femur and sternum (Fig. 2A). Magnetic resonance imaging (MRI) showed

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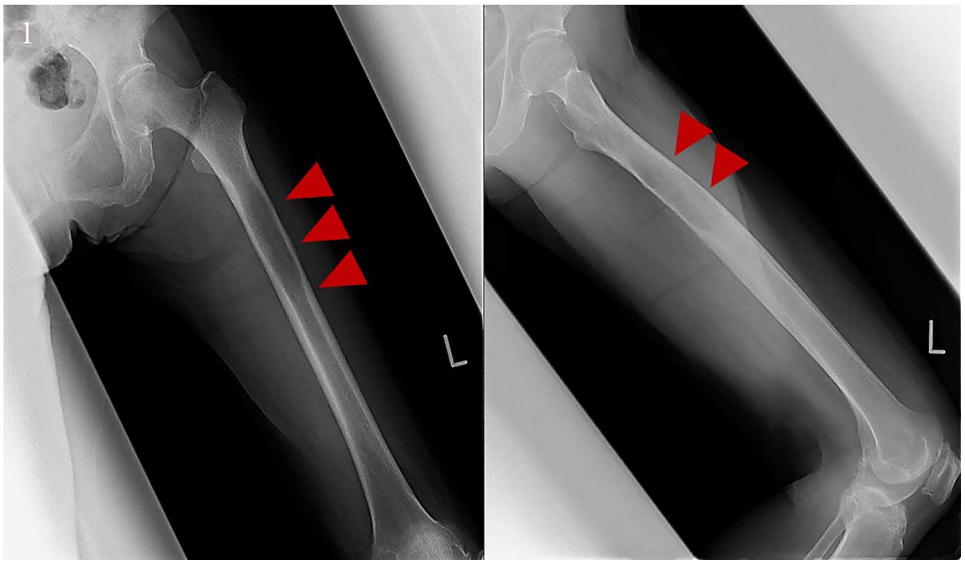

Figure 1: X-ray showed osteolytic lesion in the left femur.

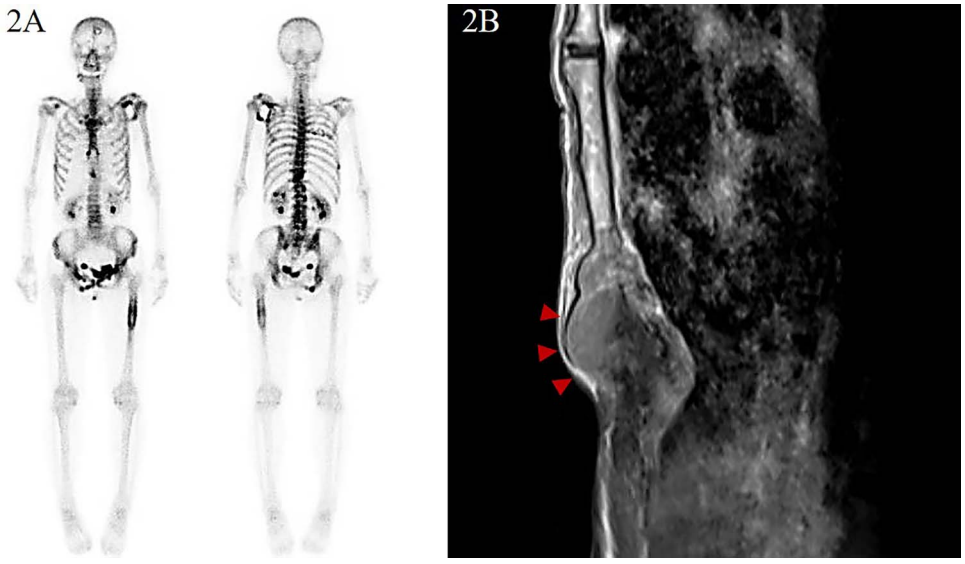

Figure 2: (A) PET-CT suspected multiple bone metastasis including frontal bone, mandible, left scapula, rib, vertebra, pubis, left femur and sternum; (B) MRI showed enhanced mass in the sternum.

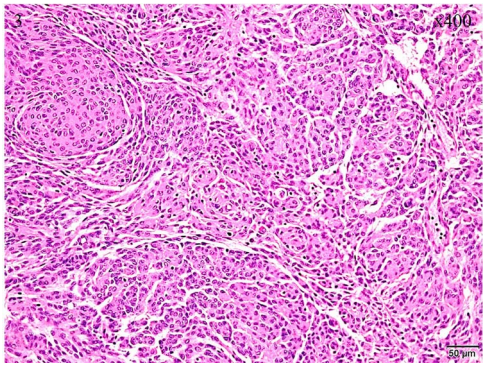

Figure 3: Tumor cells in the surgical specimen of the lung showed typical whorl formation.

enhanced mass in the sternum, which may be pricked with a needle biopsy easily (Fig. 2B). A needle biopsy of the sternum was performed. Histopathological diagnosis was consistent with metastasis of primary pulmonary meningioma, World Health Organization (WHO) grade III. Tumor cells in the surgical specimen of the lung showed typical whorl formation and positive of ecological momentary assessment (EMA) and vimentin in immunohistochemical staining (Figs 3 and 4). Tumor cells of the biopsy specimen showed malignant progression with high Ki-67 index; EMA and vimentin were also positive in immunohistochemical staining (Fig. 5). We started to treat the patient with denosumab and radiation therapy targeting the left femur. Two months after the treatment, the mass showed radiological partial response and the pain had decreased, she could walk with no cane.

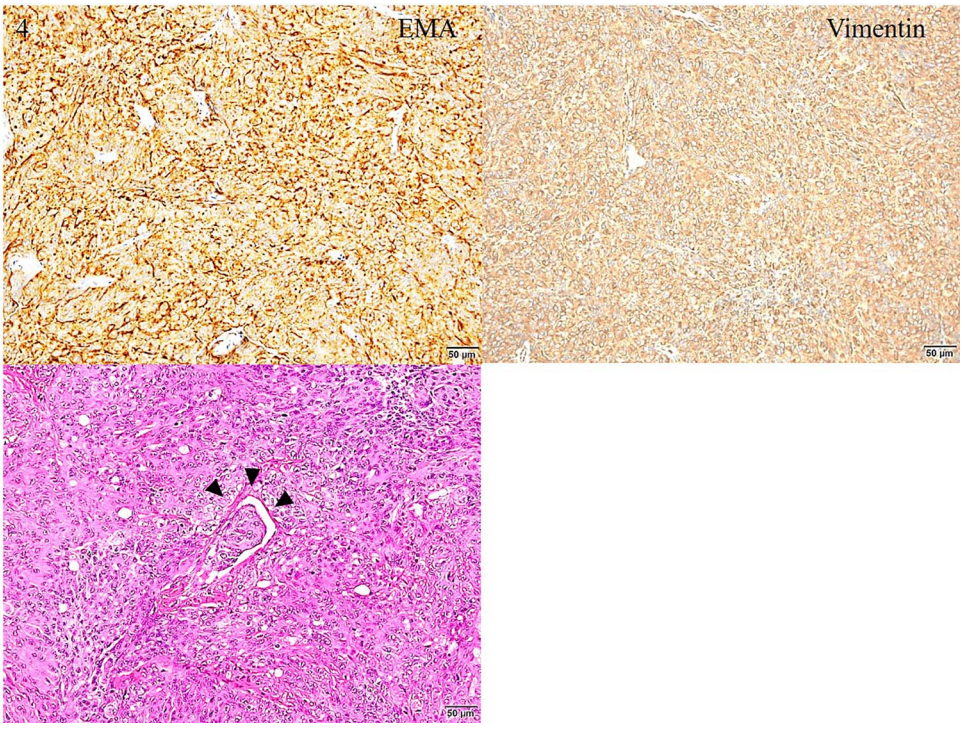

Figure 4: EMA and vimentin were positive in immunohistochemical staining. There were some aggressive vascular invasions in the surgical specimen.

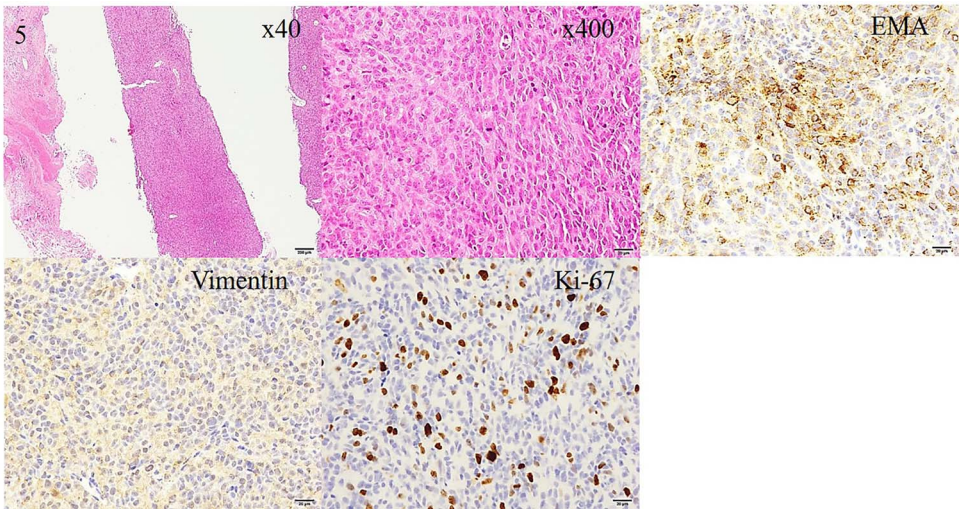

Figure 5: Tumor cells proliferated with no specific structure like sarcoma. EMA and vimentin were positive in immunohistochemical staining. Ki-67 index was partially beyond 20%. Histopathological diagnosis was metastasis of primary pulmonary meningioma, WHO grade III.

DISCUSSION

A primary pulmonary meningioma is extremely rare, but may mimic any other pulmonary tumor presenting as a solitary pulmonary nodule [7]. As far as we investigated only <45 cases have been reported so far since its first report by Kemnitz and Heinrich [4–6]. Among these cases, only five cases were malignant or atypical [1, 2]. A primary pulmonary meningioma is largely benign, grows slowly and has an excellent prognosis. Incarbone *et al.* [8] reviewed 37 cases of primary pulmonary meningiomas reported in the literature. Among 25 histologically confirmed primary pulmonary meningioma patients with radiological data of the CNS and histological assessment, 23 (92%) cases had benign and 2 (8%) cases had a malignant primary pulmonary meningioma.

Dalle Ore *et al.* [9] reviewed 1193 patients treated for meningioma including intracranial meningiomas. Ten patients had extracranial lesions suspicious for metastasis. At biopsy, eight were meningioma metastasis, one patient was a non-meningioma malignancy and the other patient was lost to follow-up prior to biopsy. Biopsy-confirmed metastasis occurred in the liver (5), lung (3), mediastinum (1) and bone (1). The observed incidence of metastasis was 0.67% ($n = 8$). Incidence increased to 2% of WHO grade II and 8.6% of grade III meningiomas. Among these patients, no case of

extracranial meningioma with bone metastasis has been reported.

We have reported rare case of malignant extracranial meningioma with multiple bone metastasis. By needle biopsy, we confirmed histopathological diagnosis as metastasis of primary pulmonary meningioma, WHO grade III. Moreover, pathology of the surgical specimen showed some vascular invasions in Fig. 4, suggesting malignant behavior. In conclusion, our experience with the present case in primary pulmonary meningioma has shown that we should think about extracranial meningioma as differential diagnosis in case of the patient with bone metastasis. The patients with extracranial meningioma should therefore be carefully monitored.

ACKNOWLEDGEMENTS

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CONFLICT OF INTEREST STATEMENT

None.

FUNDING

None.

ETHICAL APPROVAL

The patients and/or their families were informed that data from medical records would be submitted for publication and gave their consent. Ethical approval for this study was obtained from an ethic committee in our institution.

CONSENT

Written informed consent was obtained from the patients for their anonymized information to be published in this article.

GUARANTOR

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