Pulmonary and Pleural Metastases from Benign Meningeal Meningioma: A Case Report

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Meningiomas are generally benign tumors, but rarely metastasize outside of the central nervous system. A 25-year-old female was admitted to our institute because of an abnormal shadow on her chest x-ray. A computed tomography (CT) scan showed a 3-cm, well-circumscribed mass in the right lower lobe of the lung. We performed thoracotomy and resected three pulmonary tumors at the right lung and diaphragm. Histological examination revealed a benign meningothelial meningioma. Six months later, she complained of heaviness of her head and a head CT scan revealed an intracranial mass. A craniotomy was performed and a brain tumor was found to be histologically identical to the lung tumors. During the 21 years since the first operation, we performed three times of pulmonary and pleural metastasectomies and two times of resection of intracranial local recurrences. All of those tumors were meningothelial meningioma without malignant change. The patient is alive without metastasis after the last resection of metastatic tumors.

Keywords: meningioma, benign, metastasis, lung, pleura

Introduction

Meningiomas are generally benign intracranial tumors. An estimation of 0.1% metastases is reported.1) Tumors with malignant histological features have a higher rate of recurrent and metastatic lesions; however, there are few reports of metastasis from “benign” meningiomas. We present a case of meningioma with benign histology and metastases to the lung and pleura.

Case report

The patient was a non-smoking 25-year-old woman. A chest X-ray taken as part of a health examination showed a tumorous shadow in the right lower lung field and the patient was referred to our institute. She did not complain of cough, headache or any neurological symptoms. A magnetic resonance imaging (MRI) scan of the chest demonstrated a 3-cm, well-circumscribed, high intensity in T2 weighted image mass in right lung lower lobe (Fig. 1a) and three small tumors less than 1cm near the right diaphragm. There was no mediastinal lymphadenopathy.

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head. A head computed tomography (CT) scan showed an enhanced, 5-cm tumor in the right parietal lobe. The tumor removal was classified as Simpson grade IV including partial superior sagittal sinus, falx and abnormal bone. The histopathology of the tumor was a benign meningothelial meningioma. We finally diagnosed the patient as pulmonary and pleural metastasis of the intracranial benign meningioma.

Four years later, the patient exhibited a re-growth of the brain tumor and underwent a second excision. After 2 years, a follow-up chest X-ray showed an abnormal mass in the right middle lung field. A CT scan revealed four lesions at the attached vertebra and the middle lobe of the lung. A re-thoracotomy was performed and the intrapulmonary tumor was histologically identical with the brain tumor. A decade later, a follow-up MRI of the
brain showed an enlarged tumor and a third excision was done. Furthermore, one year later, a chest CT scan revealed multiple pleural tumors in the right hemi-thorax (Fig. 1b). We removed 13 tumors around the chest wall and diaphragm, which had invaded the parietal pleura and muscle. These tumors tended to be in the caudal part of the thoracic cavity. Histopathologically, pleural tumors were identical to the primary intracranial tumor. Conforming to a World Health Organization (WHO) grade I meningioma, this tumor lacked atypical features, and had a mitotic count of less than one per 10 high power fields (Fig. 2c). The patient is currently alive and has had no additional tumor recurrence in the pleural cavity 7 years after the last surgery.

Discussion and Conclusion

Meningiomas are usually non-invasive, do not metastasize and are perceived as benign tumors. Although they are typically slow growing, cranial, extra axial, benign tumors, ectopic meningiomas have been observed. In our case, the previous existence of pulmonary and pleural meningiomas remains a possibility, but it appears that the pulmonary lesion was preceded by the intracranial meningiomas. The lungs are the most common sites for metastasis of meningioma, followed by liver, bones, pleura, mediastinum, and lymph nodes.1) Our patient had repeated metastasis of meningioma to the lung and pleura. To our knowledge, this is the second case of metastasis to the pleura from benign (WHO grade I) meningioma reported in literature.2) The route of the first metastasis in our case is unclear, but may have been through the superior sagittal sinus and caval system given the parafalcine location of the primary tumors and history of prior intracranial surgery. Around the second metastasis and since then, there is a possibility that the operative procedure results in dissemination.

The histological tumor grade of the meningioma is the most important predictor for a recurrence or metastases. The recently updated 2007 WHO classification scheme divides meningiomas into three grades based on histology. More than 90% of grade III tumors exhibit aggressive behavior (local invasion and/or distant metastasis).3) WHO grade I tumors almost never metastasize or invade adjacent structures. However, as this case demonstrates, a subset of WHO grade I meningiomas can have malignant potential. One potential predictor for aggressive meningioma is the Ki-67 index, a marker of cell proliferation. The Ki-67 proliferative index (PI) has been identified as an independent predictor for both survival and tumor recurrence in meningiomas. In a retrospective study by Ohta, et al., 14 recurrent meningiomas had a mean PI of 5.7%, metastatic meningiomas had a mean PI of 14.7%, and nonrecurrent, nonmetastatic meningiomas had a mean PI of 1%.4) Our patient’s Ki-67 PI was less than 1% in the latest pleural tumor, so that, for reasons unknown, the Ki-67 PI in our case does not appear to be an accurate predictor for metastatic potential.

The discovery of metastasis often occurs after local recurrence of the primary tumor. The 5-year recurrence rate for meningiomas with benign histological features is approximately 3%.5) However, the recurrence rate is 38–78% for tumors with atypical characteristics including nuclear pleomorphism, necrosis, elevated mitotic activity, loss of whorl formation or sheeting, small cell change, macro nucleoli, brain invasion and complex karyotypes.6) Kaminski, et al. reported that tumors with clearly malignant features have a higher metastatic rate.7) Certain histological subtypes are inherently prone to recur or spread, including the WHO grade II clear cell and chordoid variants and the WHO grade III rhabdoid and papillary variants. Of these, the papillary variant carries the highest risk of late distant metastases.8,9)

Unfortunately, benign (WHO grade I) meningioma often transforms to histological malignancy.8) Our case is unique in that 20 metastatic lesions of the lung and pleura tumor were excised without histological transformation to malignancy in the course of 21 years. The possibility of metastatic meningioma should be considered in a patient with pulmonary and pleural masses and a history of an intracranial meningioma.

We diagnosed this case as histologically benign, but clinically malignant SDC because of metastasis. In such situation where a discrepancy exists between histological and clinical image, we cannot decide whether to remove the tumor or not when patients have recurrence or metastasis. Some reported that the metastatic SDC which transforms to histological malignancy has very poor prognosis, thus we should not remove the metastatic tumor.6) In our case, the patient could be alive for 21 years since the first operation because of having performed three times pulmonary and pleural metastasectomies on her. Therefore, we conclude that in order to achieve a good prognosis, we should remove metastatic tumor that do not transform to histological malignancy.
Disclosure Statement

Yujiro Nakayama and other co-authors have no conflict of interest.

References