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Case Report

Rare pulmonary metastases of atypical meningioma diagnosed on total-body ^{18}F-FDG PET/CT

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Abstract: Here we reported a 59-year-old male who had undergone brain surgery three times and the pathological results showed atypical meningioma (2015, WHO grade I; 2018, WHO grade II; 2019, WHO grade II-III), with multiple pulmonary nodules, which arose during follow-up. A total-body ^{18}F-FDG PET/CT showed multiple solid nodules with increased ^{18}F-FDG metabolism (SUVmax = 8.6). The patient underwent a CT-guided lung biopsy and the histopathological study showed positive staining of epithelial membrane antigen (EMA), vimentin (VIM), SSTR2, Ki67 (20%), and negative staining of CK, TTF-1, CD34, SY, PR, P40, respectively. Based on the history and immunohistology results, multiple pulmonary metastases from atypical meningioma were finally diagnosed, since double positive staining of EMA and VIM supported the diagnosis of meningioma and negative staining excluded primary lung cancers. The patient has given up any treatment because of personal reasons. Pulmonary metastasis from meningioma is rare, accurate diagnosis should be based on medical history, imaging characteristics, and histopathological findings.

Keywords: Pulmonary metastasis, meningioma, PET/CT, epithelial membrane antigen, vimentin

Introduction

As the most common primary intracranial tumor, most meningioma cases demonstrate benign behavior, with only 2-10% of cases behaving aggressively and 0.1-1% of cases developing distant metastases [1]. The most common target organ of metastatic meningioma is the lung, accounting for about 60% of all distant metastasis [2]. However, the clinical features of lung metastasis from meningioma vary with different cases, especially the total-body PET-CT results remain unknown. Here we describe a rare case of multiple pulmonary metastases from a recurrent meningioma, with the total-body PET-CT results reported.

Case presentation

A 59-year-old male patient was admitted to our department for multiple pulmonary nodules found by routine CT examinations. The patient underwent three times of total resection of the brain tumor in 2015, 2017, and 2019. Histological examination showed atypical meningioma (2015, WHO grade I, Ki-67 10%; 2018, WHO grade II, Ki-67 15%; 2019, WHO grade II-III, Ki-67 20%). The patient underwent no adjuvant therapies after the surgery. Multiple pulmonary lesions were found by chest X-ray in 2020, which grew during the follow-up. The CT scan showed multiple solid nodules of different sizes in both lungs, and the larger one was located in the upper lobe of the left lung, with a diameter of about 47 mm, which was considered as the metastasis of meningioma.

After the admission, a total-body ^{18}F-FDG PET/CT was conducted, showing multiple solid nodules with different sizes in both lungs (**Figure 1A**). The larger one is about 43 * 36 mm and is located in the upper lobe of the left lung. FDG metabolism is increased, SUVmax = 8.6 (**Figure 1B**, **1C**). To confirm the clinical diagnosis, we arranged a CT-guided biopsy for the patient. The immunohistochemistry result showed positive staining of epithelial membrane antigen

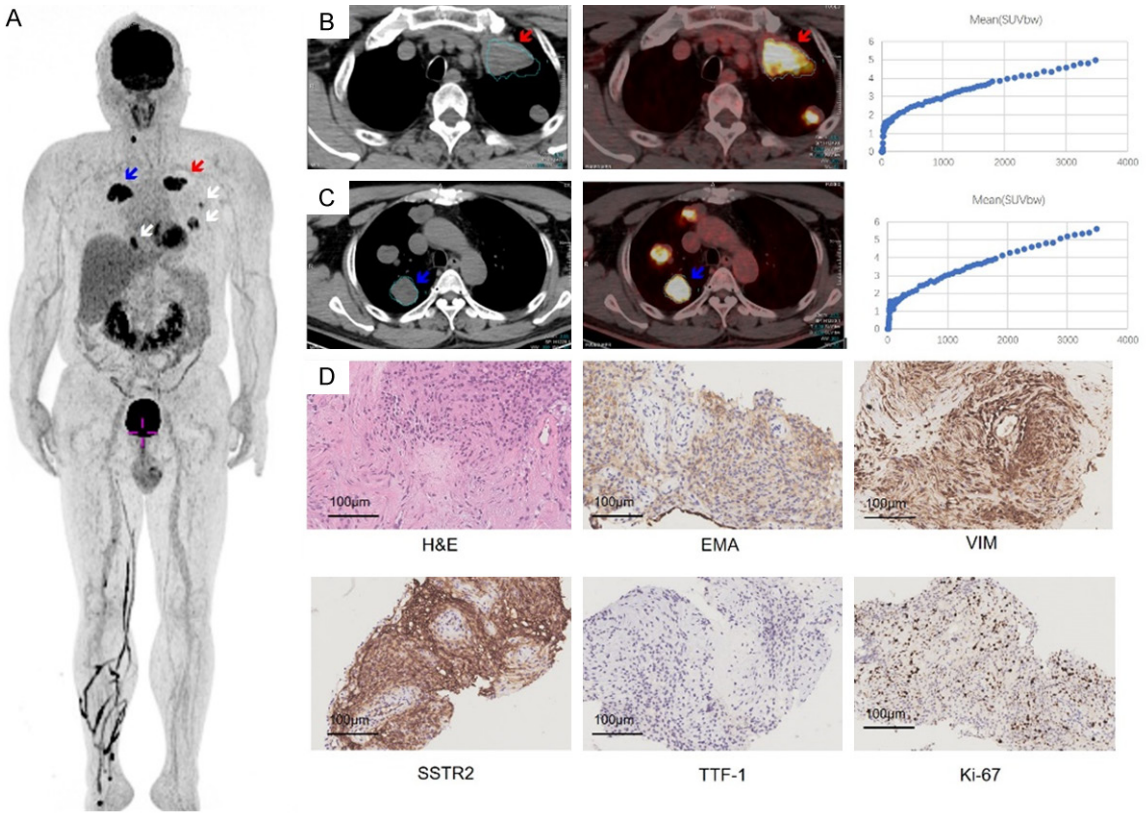

Figure 1. A total-body ^{18}F-fluorodeoxyglucose positron emission tomography/CT (^{18}F-FDG PET/CT) examination and histopathological staining results of a 59-year-old male patient diagnosed with multiple pulmonary metastases from atypical meningioma. Multiple solid nodules of different sizes were seen in bilateral lungs (A, arrow). The largest lesion of 43 * 36 mm was located in the left upper lobe with increased ^{18}F-FDG metabolism (SUVmax = 8.6) (B, arrow). Right lesions also showed time-dependent increased ^{18}F-FDG metabolism (C, arrow). Representative positive staining of epithelial membrane antigen (EMA), vimentin (VIM), SSTR2, Ki67 (20%), and negative staining of TTF-1 was revealed by CT-guided lung biopsy and the histopathological study (D).

(EMA), vimentin (VIM), SSTR2, Ki67 (20%), and negative staining of CK, TTF-1, CD34, SY, PR, and P40, respectively (**Figure 1D**). Based on the medical history, radiology, and histological results, the multiple pulmonary metastases from atypical meningioma were finally diagnosed. After the diagnosis, a following comprehensive therapy including chemotherapy and target therapy was suggested, but the patient and his family refused the following treatment because of personal reasons.

Discussion

Wang et al. reported a 59-year-old man with multiple pulmonary metastases from a recurrent intracranial meningioma and summarized a total of 35 such cases reported since 1960 [3]. They found that the median age was 50 years, while the interval between the diagnosis of the primary intracranial meningioma and the detection of pulmonary metastases ranged from 2 months to 26 years. The radiology results demonstrated that the multiple metastases were located in bilateral sites mostly, while only six cases only had right lung lesions. The age of our case was 59-year-old, and the detection of pulmonary metastases was 5 years after the first surgery and 1 year after the third surgery. The lesions in this case located in both sites of the lung.

According to 2016 WHO classification of central nervous system tumors, meningiomas are classified into grade I, grade II, and grade III [4]. In this case, the WHO grade and Ki-67 became higher and higher with the redo-surgery. In the primary surgery of 2015, the WHO grade was I and the Ki-67 was 10%, which increased into grade II and 15% in 2018, and grade II-III and

20% in 2019 respectively. The recurrence behavior of meningioma could be attributed to somatic genetic mutation [5]. He et al. found the genetic mutation including amplification in 1q and chr12; loss in 1p, 9p, and 22q; and catastrophe in chr8 and chr17 in both the previous brain meningioma and lung tissue, which validated the diagnosis of lung metastasis [2]. However, our case didn't receive the gene sequencing.

The total-body ^{18}F-FDG PET/CT also helps to make the diagnosis of multiple pulmonary metastases from meningioma, which shows the dynamic SUV curves of every lesion [6]. In this case, the PET/CT depicted the dynamic SUV curves of lesions in the lung and found no abnormal increase in FDG metabolism in the operation area. However, the final diagnosis depends on the histological results, especially the immunohistochemistry. Sathirareuangchai et al. reported that positive immunohistochemical staining with epithelial membrane antigen (EMA) and vimentin (VIM) was helpful in the diagnosis of lung metastasis from meningioma [7]. Our study also found the positive staining of epithelial membrane antigen (EMA), vimentin (VIM), SSTR2, Ki67 (20%), and negative staining of CK, TTF-1, CD34, SY, PR, P40 in the immunohistochemistry. The negative staining was used to exclude primary lung cancers.

In conclusion, here we reported a rare case of pulmonary metastasis from meningioma, of which the accurate diagnosis should be based on medical history, imaging characteristics, and histopathological findings.

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Disclosure of conflict of interest

None.

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