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Case Report

Meningioma with Extracranial Pulmonary Metastasis: A Case Report and Literature Review

Jiawei Wu[1#], Zhangqi Dou[1#], Yasaman Iranmanesh[2], Buyi Zhang[4], Yong Hou[5], Junxing Wang[1], Biao Jiang[3*], Zefeng Wang[1*], Chongran Sun[1*]

[1] Department of Neurosurgery, Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou, China

[2] School of Medicine, Zhejiang University, Hangzhou, China

[3] Department of Radiology, Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou, China

[4] Department of Pathology, Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou, China

[5]Department of Neurosurgery, Taizhou Hospital of Zhejiang province, Taizhou, China

[#]Authors Contributed Equally

[*]**Corresponding author(s):** Biao Jiang, Department of Radiology, Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou, China

Zefeng Wang, Department of Neurosurgery, Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou, China

Chongran Sun, Department of Neurosurgery, Second Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou, China

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Abstract

Meningiomas are usually benign neoplasms in which extracranial metastases occur very rarely. We report a case of an extracranial metastatic tumor diagnosed as a chordoma-like (WHO II) meningioma. A 37-year-old female presented with repeated numbness of the left limbs for two months. Enhanced cranial MRI (Magnetic Resonance Imaging) and MRV (Magnetic Resonance Venous) imaging showed a significantly enhanced tumor that invaded the superior sagittal sinus. To protect the draining vein, we performed a Simpson grade II mass removal, and there was sagittal sinus bleeding during the operation. The histopathologic finding indicated a chordoma-like meningioma. After tumor resection, there was no tumor recurrence.

However, two and a half years later, the patient started to experience substernal pain, and the chest CT scan revealed right lung nodules. To obtain a definite diagnosis, the patient underwent a CT-guided lung biopsy, and the histopathologic findings supported pulmonary metastasis of meningioma. Then, the patient underwent thoracoscopic tumor resection, and after following up the patient by telephone for approximately one and a half years after chest surgery, there was no tumor recurrence of the cerebrum or lung. In conclusion, high-grade meningiomas are prone to metastases, especially those that invade the cranial sinus. Regular follow-up must be performed. Finally, evaluations of the chest, abdomen, and bone are necessary, especially when related symptoms or signs develop.

Keywords: High-grade; Lung/pulmonary; Meningioma; Metastasis; Sinus

Introduction

Meningiomas are slow-growing benign neoplasms that constitute 14% to 19% of all primary intracranial and intraspinal tumors [1]. Although these tumors are typically benign, high-grade meningiomas may occasionally behave aggressively. However, extracranial metastases are very rare, occurring in only 0.1-0.2% of patients [2,3]. Here, we report a case of extracranial metastases to the lung from a chordoma-like (WHO II) intracranial meningioma after cranial tumor resection. Consent was obtained from patients for the retrospective review of their brain MRI, histopathological data and medical records.

Case Information

Clinical Manifestations

A 37-year-old female patient was admitted to our hospital on

September 2, 2015, due to repeated numbness of the left limbs. The paralysis of the left limbs was paroxysmal, with each episode lasting more than ten minutes and a frequency of 2-3 times a week.

Imaging Examination

Enhanced cranial MRI (Magnetic Resonance Imaging) and MRV (Magnetic Resonance Venous) imaging (Figure 1) showed that the right parietal mass, which was approximately 6.9 * 5.0 cm in size, was cystic and solid with T1 isointense and T2 hyperintense signals and that the tumor had significant enhancement. The tumor was connected to the meninges with a broad base, and a meningeal tail sign could be seen adjacent to the parenchymal edema; the right ventricle was slightly compressed. The central structure slightly deviated to the left side. The tumor on the right parietal lobe invaded the superior sagittal sinus, the boundary was unclear, a filling defect could be seen behind the posterior superior sagittal sinus, with multiple surrounding collateral vessel shadows. No abnormal signals were found in the bilateral sinus cavities, transverse sinus, and sigmoid sinus. Diagnosis: Meningioma was considered, but hemangioma was not entirely excluded. The tumor on the right parietal lobe invaded the superior sagittal sinus.

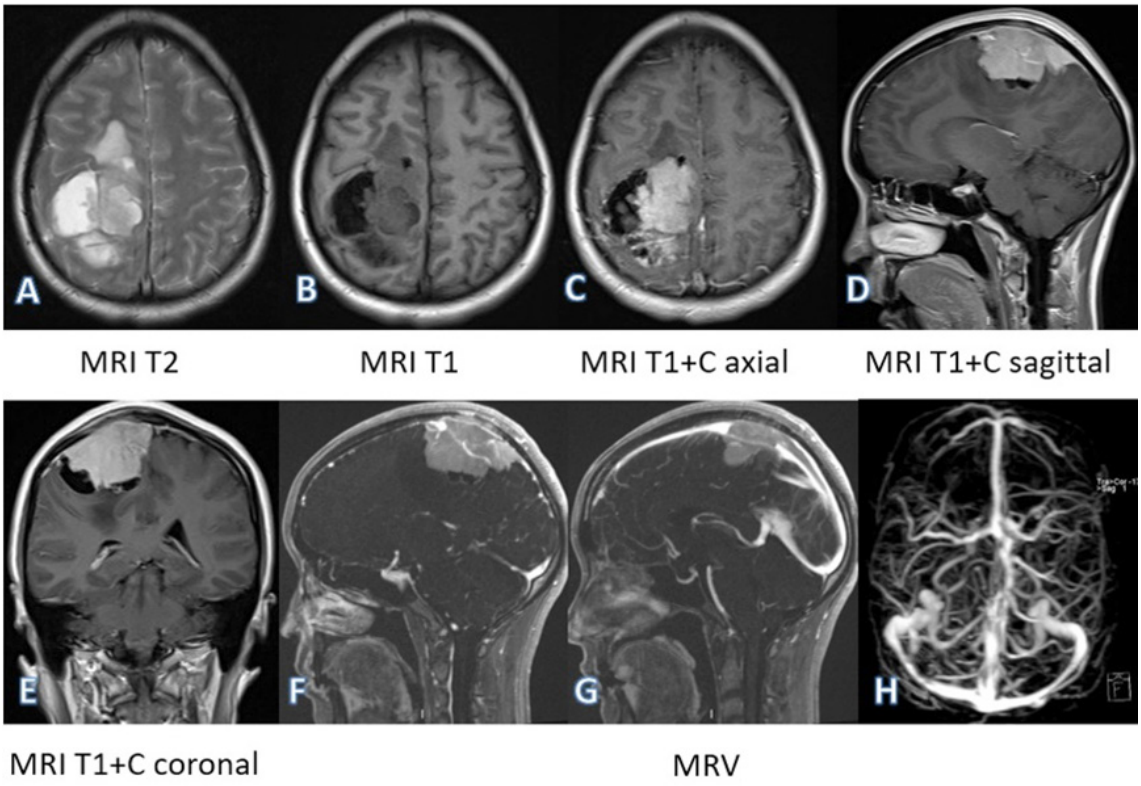

Figure 1: A and **B**: the right parietal mass, which was approximately 6.9 * 5.0 cm in size, was cystic and solid with T1 isointense and T2 hyperintense signals; **C-E**: the tumor had significant enhancement; **F-H**: The tumor on the right parietal lobe invaded the superior sagittal sinus, the boundary was unclear, a filling defect could be seen behind the posterior superior sagittal sinus, with multiple surrounding collateral vessel shadows.

Surgical Treatment

A right parietal midline incision was made. During the operation, a tumor was located beneath the meninges, showing a pale yellow color, a tough texture, a size of approximately 7 * 7.5 cm, and abundant blood supply. The base was located on the convex surface of the meninges, the sickle of the brain, and the sagittal sinus wall. Obvious adhesions to the brain tissue were observed. The tumor was mainly removed under a microscope. A thick vein passed from the deep part of the tumor and extended into the sagittal sinus along with the sickle of the brain. To protect the draining vein, a small amount of tumor was retained. After the resection, there was sagittal sinus bleeding, which was caused by the reconstruction of the sagittal sinus using gelfoam and BioGlue. Simpson grade II tumor resection was achieved. Intraoperative frozen pathology results showed meningioma with atypical cells and dense cells in some areas. After the

operation, the patient was in good general health, and the numbness of the left limbs did not recur. Cranial CT and MRI showed no abnormalities one week postoperation.

Histopathologic Diagnosis

Postoperative pathology revealed right parietal meningioma with atypical cells, interstitial mucinous degeneration, and occasional mitosis, which was considered to be chordoma-like (hematoxylin-eosin, ×400). The immunohistochemical results were as follows: Ki-67 5%, P53 +, EMA +, GFAP-, NSE foci +, CD99-, CD56 foci +, and CK (AE1/AE3) (Figure 2).

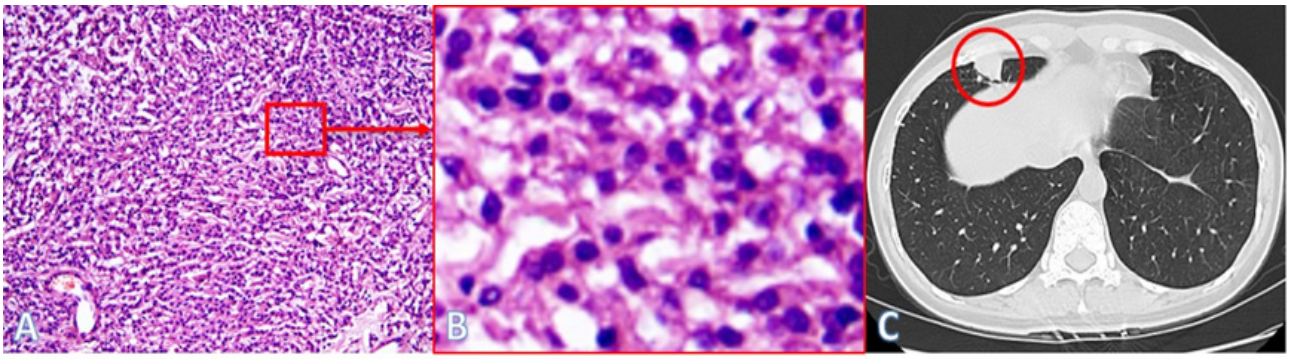

Figure 2: A and **B**: right parietal meningioma with atypical cells, interstitial mucinous degeneration, and occasional mitosis, which was considered to be chordoma-like (hematoxylin-eosin, ×40). The immunohistochemical results were as follows: Ki-67 5%, P53 +, EMA +, GFAP-, NSE foci +, CD99-, CD56 foci +, and CK (AE1/AE3); **C**: right middle lobe nodules with a maximum diameter of 20mm shadow.

Postoperative Follow-up

Follow-up images of enhanced cranial MRI within four years after surgery (Figure 3).

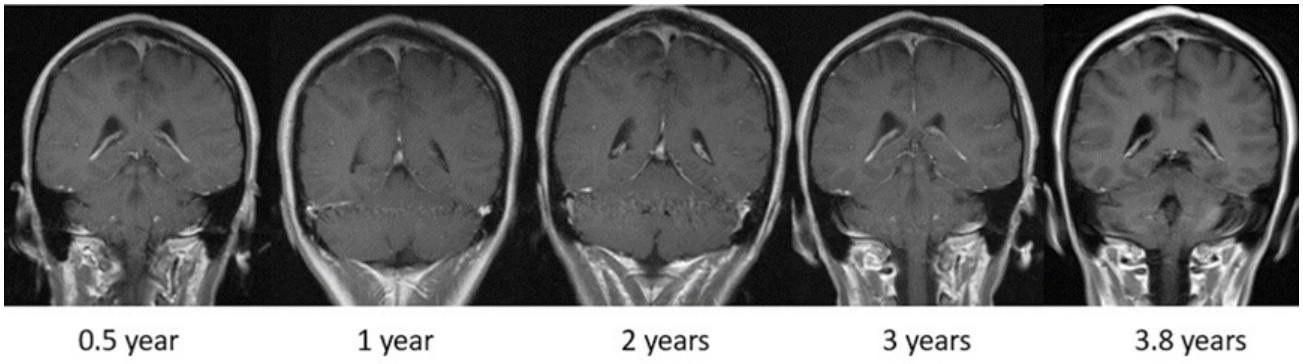

Figure 3: Enhanced cranial MRI was performed after surgery with no visible abnormal changes.

Pulmonary Metastases

Two and a half years after tumor resection, the patient presented with substernal pain without any apparent inducing factors. The chest pain was described as paroxysmal dull pain and was exacerbated with deep breaths. There were no other simultaneous symptoms, such as cough, phlegm, belching or acid regurgitation. The chest CT scan revealed right lung nodule (Figure 2). However, no significant improvement was observed after anti-inflammatory treatment. To obtain a definite diagnosis, CT-guided lung biopsy was performed in our hospital. A total of two gray-white lesions were removed. The histopathologic findings supported pulmonary metastasis of meningioma. After undergoing puncture biopsy in our hospital, the patient underwent thoracoscopic tumor resection in another hospital. The postoperative histopathological report supported the histopathologic description of the puncture biopsy results in our hospital. After following up the patient for approximately one and a half years by telephone, the general medical condition of the patient was acceptable (KPS score 100 points). The patient had no head discomfort, seizures, numbness in the left limbs, or significant chest discomfort.

Discussion

Meningiomas are primary Central Nervous System (CNS) tumors that originate from arachnoid cap cells around the brain [4]. At present, meningioma has surpassed glioma as the most common primary central nervous system tumor [5]. The vast majority of meningiomas are considered benign and can be treated with surgical resection and adjuvant radiation therapy. Compared to other central nervous system tumors, extracranial metastatic meningiomas are extremely rare, occurring in only 0.1-0.2% of patients [2,3]. The organs most likely to develop metastases are the lung (37%), bones (16.5%), spine (15.2%), liver (9.2%), adrenal glands, neck, and other structures (21.9%) [6-11]. Atypical or anaplastic meningioma is prone to extracranial metastasis [12].

According to the 2016 edition of the World Health Organization (WHO) classification of neurological tumors (CNS), the histopathologic classification of 15 meningiomas is listed as follows [13]. The subtypes that are prone to extracranial metastasis are papillary meningioma (WHO II), atypical meningiomas (WHO II), and anaplastic meningiomas (WHO III) [14-16]. Certain imaging features, such as mushroom-like growth, non-uniform enhancement, peritumoral edema, osteolysis, inherent cyst-like areas, and fuzzy tumor-brain borders, are considered to be important clues for the diagnosis of malignant or aggressive tumors [17]. Furthermore, immunohistochemical analysis of nuclear protein, Ki-67 and P53 marker indexes related to cell proliferation or molecular markers such as CDKN2A deletion and 9p21 deletion are very useful for assessing the possibility of tumor recurrence or metastasis [18,19].

The Ki-67 index is significantly elevated in benign meningiomas (mean 3.8%), atypical meningiomas (mean 7.2%) and anaplastic meningiomas (mean 14.7%) [20]. Intracranial lesions usually recur several times locally before metastasis, but in this case, no intracranial recurrence occurred before extracranial metastases [3]. One of the most common chromosomal aberrations of meningiomas is the partial or complete deletion of chromosomes. Somatic mutations of the NF2 gene at 22q12.2 have been described as early events of tumorigenesis [21]. The second most significant genetic change of meningiomas is the partial or complete deletion of 1p, which is also the most common progression-related chromosomal aberration in meningiomas [22]. In related studies, mutations of 22q and 1p in primary intracranial meningiomas and lung metastases were detected. The chromosomal changes in the latter may reflect the chromosomal and genetic heterogeneity of these tumors [23].

The location of meningioma metastases mainly depends on the route of spread. Four different routes of transmission have been proposed [24]: (i) transjugular vein (supported by evidence of cervical lymph node, thyroid, lung/pleural metastasis); (ii) paravertebral venous plexus ("Batterson's venous drainage"); (iii) tumor invasion and proximity to the vena cava; and (iv) localized metastasis via the lymphatic and cerebrospinal fluid pathways. Among these routes, hematogenous metastasis is mainly caused by tumor cells that invade the venous system and spread through the vena cava system, which may be the reason why the lung and liver are prone to metastasis [25]. Although meningiomas originate from arachnoid cells and are naturally exposed to cerebrospinal fluid during their growth or surgical intervention, metastasis through the cerebrospinal fluid is less common than hematogenous metastases to the internal and external organs [26].

The factors affecting extracranial metastasis include the following: (i) an increase in the degree of malignancy, (ii) tumor recurrence, and (iii) tumor invasion of the venous sinuses [23]. More than 90% of cases of extracranial metastasis reported in related studies occur after resection or shunt surgery. These two processes help tumor cells enter the extrameningeal blood vessels and lymphatic vessels [27]. In this case, the tumor had an abundant blood supply, with a large drainage vein on the brain surface at the anterior margin of the tumor and a vein at the posterior margin that ran deep through the tumor, which might have provided a functional pathway for tumor metastasis. There was also hemorrhage of the sagittal sinus during tumor resection, which might have caused tumor colonization in the lungs along the venous system.

The incidence of extracranial metastases caused by meningiomas is very low, and there is currently no standard treatment for extracranial metastases; however, general surgical resection is preferred [28]. Postoperative radiotherapy has been recommended to prevent local recurrence, especially when resection or histology suggest a malignant tumor [29]. In most published cases of pulmonary metastases, early complete lung tumor resection is the preferred treatment [30-34]. In this case, after the lung tumor was removed, no chemoradiotherapy was performed, and no recurrence of the lung lesions was found, which may be related to the relatively low tumor invasiveness. The prognosis of intracranial and extracranial meningioma metastases is unknown [35].

Conclusion

When meningiomas metastasize or radiologic findings indicate aggressive behavior, careful histopathological examination with immunohistochemistry analyses should be performed to determine the nature of the tumor. In intracranial high-grade meningioma, extracranial metastasis should be considered, especially when the meningioma invades the cranial sinus; regular follow-up is necessary with evaluations of the chest, abdomen and bone, especially when related symptoms or signs also develop.

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Conflict of Interest

The authors declared no conflict of interest.

Authorship

Research design: CS and ZW; data collection: Yasaman Iranmanesh, BZ and BJ; data analysis: JW (Jiawei Wu), ZD, JW (Junxing Wang) and YH; manuscript writing and revising: JW (Jiawei Wu), ZD and CS. All authors listed above contributed greatly to drafting the manuscript and performing critical revision, and had access to the data and approved the final manuscript for submission. We declare that the work described in this research paper has not been published previously nor been under consideration for publication elsewhere.

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