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CASE REPORT

Recurrent Anaplastic Meningioma with Metastasis

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ABSTRACT

Meningioma is the commonest primary intracranial tumor in adults, with the majority being benign. Malignant meningioma is exceedingly rare and carries a poor prognosis because of its tendency for local recurrence and distant metastasis. We present the case of a 53 years old male with anaplastic malignant meningioma, which recurred after excision and radiotherapy and later metastasised to pleura, bones and brain.

Keywords: Metastatic meningioma, Anaplastic meningioma, Pleural metastasis, Intracranial tumor, Extracranial metastasis

BACKGROUND

Meningioma is the commonest primary brain tumor in adults, constituting 30% of all intracranial tumors. [1-3] These arise from arachnoid cap cells located in arachnoid villi. The majority of meningiomas originate from brain meninges, but 10% of all cases originate from spinal cord meninges. [4] There are 3 subtypes of meningioma according to World Health Organization (WHO). WHO Grade 1, benign meningioma making approximately 90% of all meningiomas. WHO Grade 2, atypical meningioma represents 5-7% and WHO Grade 3, anaplastic, or malignant meningioma represent 1-3%. [1,2] Grade 3 meningiomas behave aggressively with the worst prognosis. They have a high local recurrence rate and rarely develop distant metastasis. [4,5] As extracranial metastasis is extremely rare, seen in less than 0.1-0.2 % of all meningiomas, [2,3] a high index of suspicion should be kept when encountered with such lesions in patients with recurrent atypical/anaplastic meningiomas.

CASE REPORT

A 53-year-old male had previously had a meningioma excision in 2005, but it recurred after two years without evidence of distant metastases. The first time he presented at our trust in April 2021, the MRI head revealed that the left parietal meningioma had grown in size since previous MRI imaging from 2007 to 2009, with increased surrounding vasogenic edema and mass effect as well (Figure 1).

The patient underwent a craniotomy with complete resection of the left parietal lobe lesion (histopathology confirmed anaplastic meningioma), followed by adjuvant radiotherapy in September 2021. A recurrence of the mass was detected again in June 2022, which was treated with stereotactic radiosurgery. During this period, after experiencing acute shortness of breath, the patient went to the emergency department, where a radiograph revealed a left sided pleural effusion. As a result of these findings, a chest CT scan was obtained which revealed gross left pleural effusion with enhancing nodular pleural masses bilaterally, but predominantly in the left hemithorax, with a subcarinal lymph node (Figure 2). A preliminary suspicion of mesothelioma was raised due to nodular pleural masses and pleural biopsy was performed for confirmation. Immunohistochemistry of the pleural biopsy, however, confirmed it to be a metastatic meningioma. Tumor cells showed strong diffuse positivity for EMA and strong but focal positivity for AE1/AE3.

This patient also had a CT examination of the abdomen and pelvis, which revealed a lytic lesion in the left ischium with a small soft tissue component (Figure 3). Follow up CT scans showed progression of the pleural deposits and ischial lesion. Latest MRI in January 2022 also showed a new small enhancing lesion in left temporal lobe (Figure 4). The brain and ischial lesions are not histopathologically confirmed, but given the biopsy-proven metastatic pleural deposits, these lesions are highly likely to represent metastases from meningioma. The patient's clinical symptoms deteriorated rapidly and he succumbed to disease progression in January 2023.

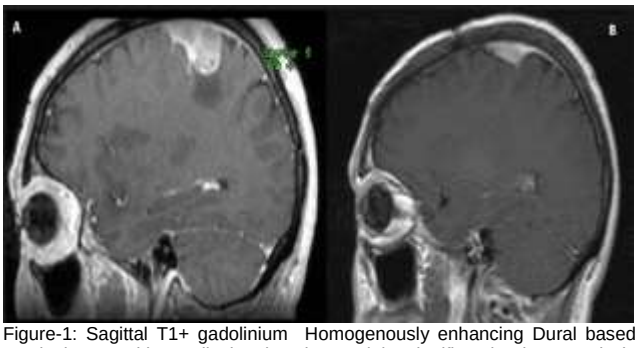

Figure-1: Sagittal T1+ gadolinium Homogenously enhancing Dural based meningioma with perilesional edema (A) significantly increased in comparison to previous MR examination (B)

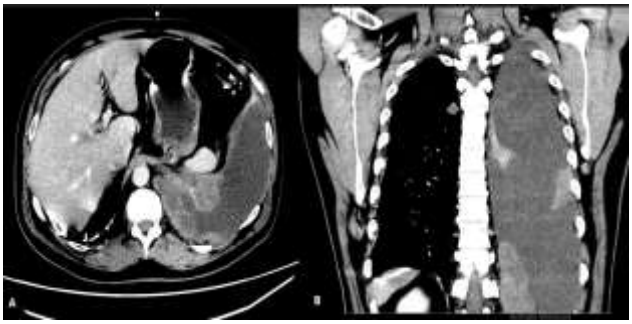

Figure-2: Contrast enhanced CT chest axial (A) and coronal (B) images showing heterogeneously enhancing nodular pleural thickening consistent with biopsy proven meningioma mets

Figure-3: Expansile lytic lesion in left ischium with soft tissue component

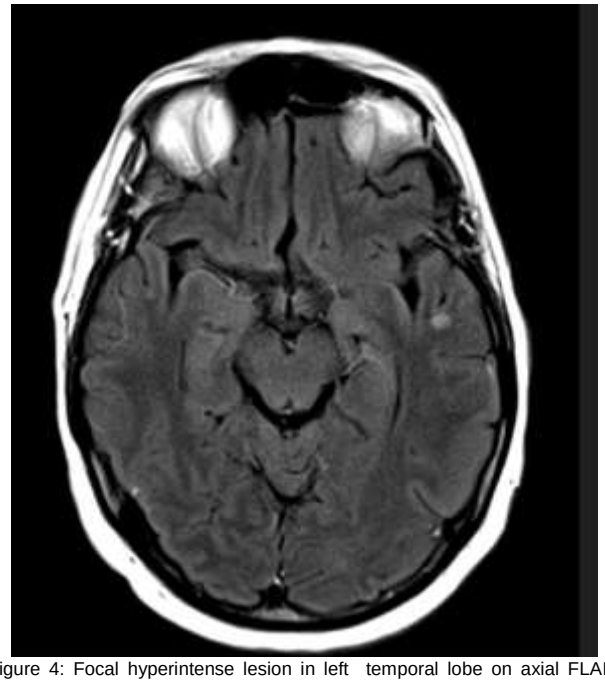

Figure 4: Focal hyperintense lesion in left temporal lobe on axial FLAIR sequence

DISCUSSION

Grade 3 meningiomas represent 1-3% of all meningiomas, yet carry the worst prognosis with 10-year progression-free survival rate of 0% and 10-year overall survival rate ranging from 14-34%. [4] Some studies report a metastatic rate of as high as 43% with Grade 3 tumors relative to 0.1 % seen in all meningiomas. [4] When metastasis do arise, most frequently involved sites include lung (37.2%), bones (16.5%), intraspinal (15.2%) and the liver (9.2%). [1,4] Pulmonary metastasis manifest mostly as multiple masses rather than as a solitary mass [2], as was the scenario in our case.

The mechanism of developing metastasis is not clearly understood, but it may be attributed to hematogenous spread or even spread via the cerebrospinal fluid. [1,2] Metastatic meningioma although a rare entity but is well documented and the metastasis may present as any other metastasis or even as a primary tumor of the site involved. Hence, it is important to keep a high index of suspicion especially in patients with recurrent and high grade meningiomas, to reach the correct diagnosis. [5] One study has even recommended systemic imaging in patients with recurrent tumors or concerning symptoms to detect extracranial metastasis and help in clinical decision making. [6]

First line treatment for Grade 3 meningiomas is complete surgical resection followed by radiotherapy to minimize the chances of recurrence, [3,4] nevertheless despite complete surgical resection, the tumor does recur in 50-90% of cases. [3] Because of this high recurrence rate, close monitoring with MRI is recommended. Once the tumor recurs locally or metastasizes, no standard treatment guidelines have been established or agreed upon. [2,3,7] However, multiple treatment strategies have been used with pulmonary metastasis, including surgical resection, adjuvant radiotherapy, chemotherapy and even cytoreductive surgery and hyperthermic intrathoracic chemotherapy. [7]

Summary: Our case is a 53-year-old male whose meningioma recurred and transformed into a higher grade 16 years after initial resection. Despite a complete surgical excision and radiation therapy as per standard treatment guidelines, the tumor recurred after 9 months of treatment. Within 3 months of the 2nd recurrence, the patient developed with pulmonary and bone metastasis and later brain deposit. Unfortunately, the patient survived 16 months after surgical resection and radiotherapy, with a progression free period of 9 months only.

Learning Points

- Malignant meningioma is exceedingly rare and carries a poor prognosis because of its tendency for local recurrence and distant metastasis.
- As extracranial metastasis is extremely rare, seen in less than 0.1-0.2 % of all meningiomas, a high index of suspicion should be kept when encountered with such lesions in patients with recurrent atypical/anaplastic meningiomas.
- When metastasis do arise, most frequently involved sites include lung, bones, intraspinal and the liver. Pulmonary metastasis manifest mostly as multiple masses rather than as a solitary mass.

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