CASE REPORT

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Disseminated Anaplastic Meningioma Before Local Recurrence

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ABSTRACT Meningiomas has account for 30% of all primary intracranial neoplasms. Extracranial meningioma metastasis is extremely rare. A 53-year-old female admitted with amnesia and headache. Subtotal resection was obtained. Tumor histopathology revealed an anapylastic meningioma and the patient received postoperative radiotherapy. Three months later, the patient experienced weakness and bone pain. Cranial MRI excluded local recurrence. Computed tomography revealed liver, bone and subcutaneous metastases. Liver biopsy confirmed the diagnosis of grade 3 meningioma. After receiving chemotherapy, CT showed progression in liver lesions and cranial MRI revealed local recurrence. The patient died 12 months after the primary surgery.

Keywords: Radiotherapy; meningioma; neoplasm metastasis

Meningiomas arise from the meningothelial cells of the dura and has an indolent nature. They account for 30% of all primary intracranial neoplasms.[1] Histological grading is based on the WHO classification and majority of them is benign (WHO grade I) with a relatively good prognosis. Grade 2 or atypical meningiomas approximately are 20% and have a higher rate of recurrence. WHO grade 3 or anaplastic variants are less than 5%, typically aggressive and have high mitotic rate.[1,2]

Extracranial meningioma metastasis (EMM) is extremely rare, occurs in 0.1-0.2% of meningioma, and may be left undiagnosed.[2,3] The most common site of metastasis is the lung, followed by the osseous involvement of long bones and the vertebral column, spinal cord and liver.[3]

EMM is more common with atypical or anaplastic histopathologies.[4-6] Multiple extracranial metastases are extremely rare.[7] No standard of care has been established for management until now and these patients have poor prognosis.[8,9]

In this article, we report a case of anaplastic meningioma metastases to in multiple sites including, liver, bone and subcutaneous nodules before local recurrence.

CASE REPORT

A 53-year-old woman was admitted with amnesia and frontal headache which had lasted for 2 months. A computed tomography (CT) of the head revealed well-defined, irregular lobulated lesion, measuring 6x5 cm in diameter which located in left frontoparietal area. The tumour was hyperdense to normal brain with intratumoural necrosis and an extensive oedema. Further magnetic resonance imaging (MRI) of the brain confirmed the lesion with contrast enhancement. Neurological examination was intact, with no cranial nerve abnormality, consciousness or cognitive deficit.

A left parasagittal craniotomy was performed and subtotal resection was obtained (Figure 1). Hematoxylin and eosin staining showed a pleomorphic, highly proliferative meningeal tumour with an overall high cell density, nuclear atypia as well as areas of necrosis, and numerous mitoses, althogether more than 10BBA in 30 hpf. Immunohistochemical staining with Ki 67 revealed a very high rate of proliferation with a rate of 80% within the tumour tissue, marking 80% most of the meningeal tumour cells displayed immunpositivity for epithelial membrane antigen and S100. These findings were consistent with an anaplastic meningioma with increased mitotic activity (Figures 2, 3, 4).

Postoperatively, the patient had no new neurological deficit and 39x36x22 mm residual tumour was detected in postoperative MRI. She was discharged on the 7th postoperative day with a good performance status. The patient received postoperative postoperative intensity-modulated and image-guided radiation therapy (IM /IGRT) with a cumulative dose of 60 Gy.

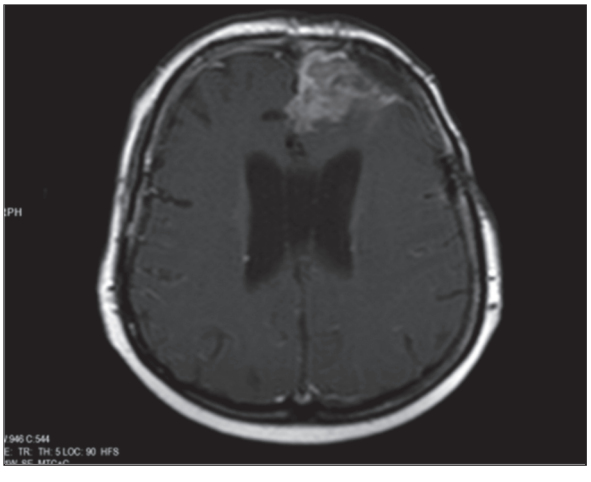

FIGURE 1: MR shows left craniectomy defect and subtotal resection area.

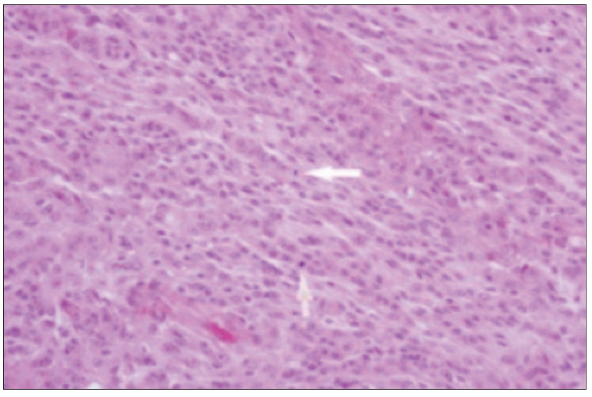

FIGURE 2: The tumor is composed of meningothelial cells characterized by round oval nucleated cells with eosinophilic syngeneic cytoplasm. Certain areas have marked cellular atypia. Frequent mitosis is observed (white arrow) (HE, x400).

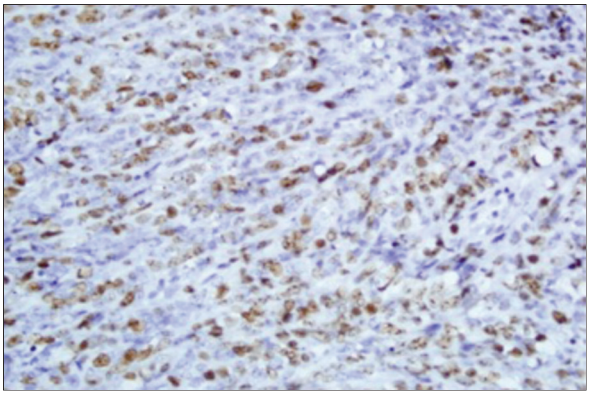

FIGURE 3: The Ki-67 proliferation index was observed above 50% (Ki-67, x400).

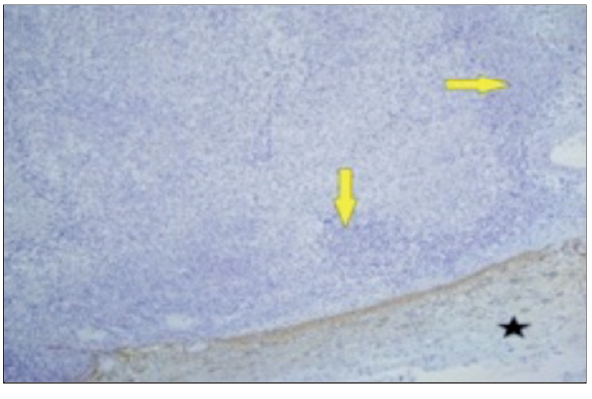

FIGURE 4: GFAP (glial fibrillary acidic protein) only stained the brain parenchyma (stellate) and no tumor staining was observed. Also present in the tumor is extensive necrosis (yellow arrows) (GFAP, x100).

Three months later, the patient experienced weakness and bone pain localized in right femur and left iliac area. Cranial MRI excluded local recurrence. Computed tomography scan of the chest and upper abdomen revealed multiple liver lesions located in the right side, multiple bone metastases and enlarged lymph nodes without chest lesion (Figure 5). A fluorodeoxyglucose (FDG) positron emission tomography (PET) CT scan showed FDG uptake in predefined CT sites (Figure 6).

Liver biopsy was performed and pathological examination of the resection specimen confirmed the diagnosis of a grade 3 meningioma similar histopathology with primary.

After receiving 5 cycles of cisplatin and etoposide, control abdomen CT showed progression in liver lesions. Meanwhile, she described frontal headache with similar to initial complaint. Magnetic resonance imaging of the brain revealed recurrence lesion at postoperative area that was approximately 55x45 mm in size. The patient refused any further treatment and was received palliative pain management.

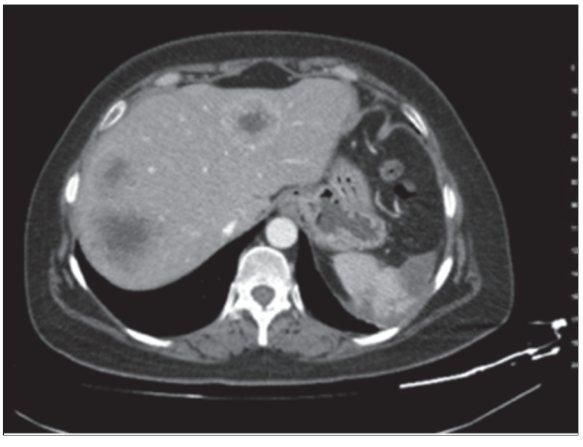

FIGURE 5: An enhanced computed tomography scan of the abdomen demonstrating multiple metastasis.

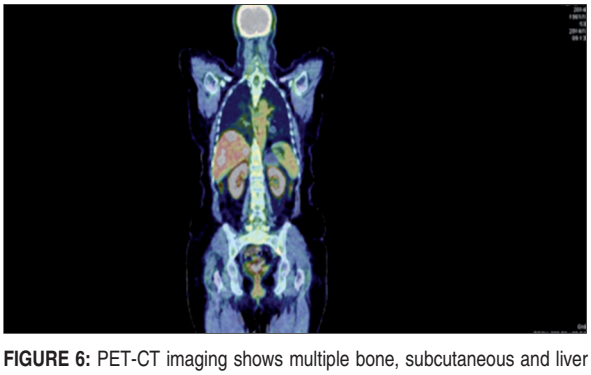

FIGURE 6: PET-CT imaging shows multiple bone, subcutaneous and liver metastasis.

The patient died 12 months after the primary intracranial surgery. No autopsy was performed.

DISCUSSION

Meningioma prevalence is 97.5 per 100,000 based on surgical series in the United States and approximately 8000 meningiomas are identified every year. In a recent epidemiologic study, they are found to be the most frequent primary intracranial neoplasm.[1]

After total resection, locoregional recurrence rates vary from 9% to 32%.[9] EMM is rare and has been estimated to occur in less than 0.1% with WHO grade 2 and 3 patients.[10] WHO grade II and grade III meningiomas are associated with poor prognosis, higher locoregional recurrence rates and metastases.[2,11]In this case report, patient had an initial diagnosis of anaplastic meningioma (WHO grade II).

According to current studies, the lungs (37.2%), are the most common site of metastasis, followed by bone (16.5%), spinal cord (15.2%) and liver (9.2%). In addition, metastases rarely occur in the kidney, bladder, thyroid, or breast.[2,3]

Pulmonary metastases typically present as single or multiple non-calcified parenchymal nodules without symptoms.[12]

Metastases are frequently diagnosed after local recurrence. Time to distant metastasis can vary from a few months to several years.[3] The mean interval between diagnosis of the primary tumour and EMM is ~6 years, the longest being 24 years.

Associated preidentified risk factors for EMM were, local recurrence, high mitotic activity, nuclear pleomorphism, previous craniotomy, venous sinus invasion, papillary morphology, hypercellularity, cellular heterogeneity.[12] Analyses of immunohistochemistry such as the Ki-67 labelling index is useful in defining aggressiveness for recurrence and metastasis.[2,7] Recent work has also highlighted several molecular and cytogenetic markers that may predict EMM such as CD90. CD90 a protein associated with aberrant activation

of the self-renewal machinery. CD90 has been previously observed in glioblastomas and may play a role in the formation of tumour vasculature and tumour progression. Scognamiglio and colleagues reported that high expression of CD90 in cases of EMM is a potential marker for metastatic potential.[6] Additionally, Frydrychowicz and colleagues suggested that deletions affecting chromosomes 22 and 1 in primary atypical meningiomas may be associated with metastasis and aggressive progress.[13]

Our patient had most of the identified risk factors for EMM with high mitotic activity, high Ki-67 index and anaplastic histology.

The exact route of metastasis is unknown, haematogenous, lymphatic, and cerebrospinal fluid (CSF) routes. CSF spread may have associated for intraspinal EMM and haematogenous spread for vertebral EMM however it is difficult to confirm.

Complete surgical resection is the treatment of choice for resectable intracranial or intraspinal meningiomas. Adjuvant postoperative radiation therapy has been recommended for the prevention of local recurrence.[6] There are some reports indicating good results obtained with chemotherapy however, traditional chemotherapy has little benefit in malignant meningioma.[14]

In conclusion, distant metastasis of meningioma is extremely rare and it usually seen after local recurrence. When local recurrence occurs or systemic sign and symptoms develop, further screening must be performed to exclude EMM.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Opinion/Concept: *İpek Pınar Aral, Mine Şalk|* ***Design:*** *Süheyla Aytaç Arslan;* ***Supervision/Consultancy:*** *Süheyla Aytaç Arslan;* ***Data Collection and/or Processing:*** *İpek Pınar Aral;* ***Analysis and / or Comment:*** *Süheyla Aytaç Arslan;* ***Resource Scanning:*** *İpek Pınar Aral;* ***Combination Writing:*** *Süheyla Aytaç Arslan, İpek Pınar Aral;* ***Critical Investigation:*** *Süheyla Aytaç Arslan, Nuran Sungu;* ***Materials:*** *Nuran Sungu.*

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