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A Rare Liver Tumor: Case Report and Literature Review

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ABSTRACT

The incidence of distant metastasis from meningioma is reported to be rare. Only about 0.1% of meningiomas metastasize outside the central nervous system (1), with 11 cases of liver metastasis reported in PubMed. A consensus about screening programs and treatment plan for metastatic meningioma is still lacking. Surgical resection remains the first option for the treatment and should be performed in Centers with adequate experience. We report a case of metastatic liver tumor from an atypical meningioma and reviewed similar previously publishedcases, summarizing potential risk factors for liver metastasis and screening recommendations.

Key words: metastatic meningioma, liver metastasis, locally recurrent meningioma, ultrasound, Somatostatin Receptor Scintigraphy

INTRODUCTION

Meningiomas are the most frequent primary brain tumors. They arise from the meningothelial cells of the arachnoid membrane and are more common in women and in the sixth decade of life (1). Most are benign, but a minority are atypical or malignant (3). Surgery and radiotherapy are the main therapeutic options for primary meningioma.

Distant metastasis from meningioma are rare. Extracranial metastases of atypical meningiomas have an estimated incidence of 1–5 out of 1000 cases, most commonly to the lung, liver, lymph nodes, bone, pleura and mediastinum (1). The proposed dissemination pathways for hepatic metastasis are the spread through the vertebral venous system to the liver (4). Without an established screening strategy, metastases are easily overlooked because most patients remain asymptomatic until an advanced stage. Therefore, regular follow-up examination appears necessary.

CASE PRESENTATION AND LITERATURE REVIEW

A 48-year-old female was diagnosed with meningioma in 2000 and underwent complete surgical resection with pathology indicating a fibrous meningioma.

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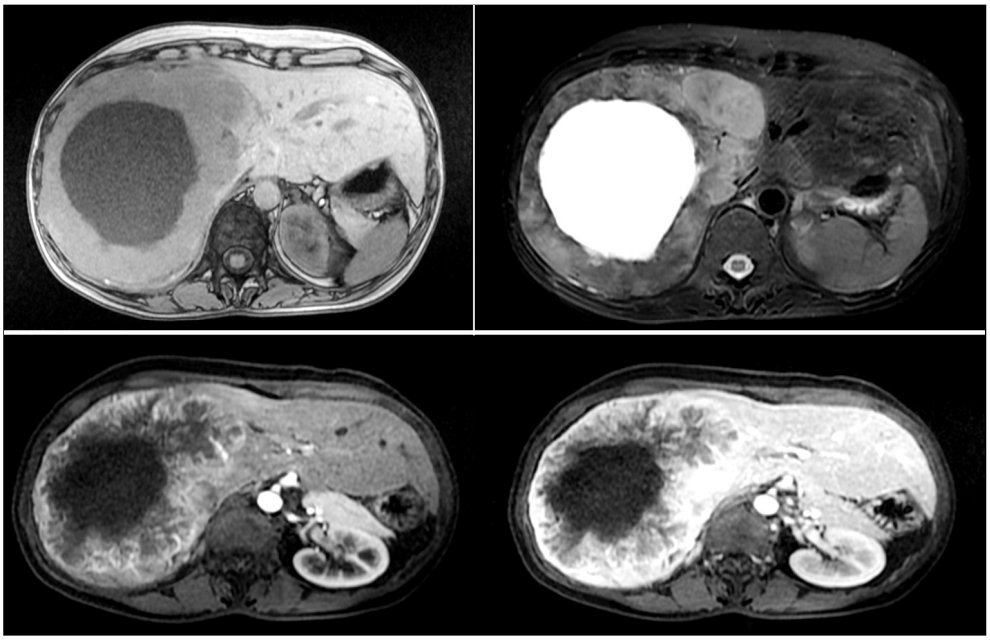

Figure 1 - MRI: the tumor shows long T1 T2 signal with restricted diffusion and heterogeneous enhancement after contrast administration

In 2008, the patient presented with blurred vision and right-sided weakness with poor coordination. Cranial computed tomography (CT) showed tumor relapse, and the patient underwent a second complete surgical resection. Pathology revealed atypical meningioma and she received post-operative radiotherapy, with regular neurosurgical follow-up.

In January 2020, the patient complained of frequent loose stools and weight loss for 6 months. Physical examination was relevant fora 5cm, hard, non- tender, hepatomegaly. Abdominal ultrasound found a large solid-cystic liver mass with internal vascularity. Differential diagnosis at the time included primary or metastatic liver neoplasm. Laboratory tests revealed normal liver function and coagulation time, negative hepatitis serology, normal tumor markers. Abdominal magnetic resonance imaging (MRI) revealed a 12 cm x 16 cm x 20 cm cystic-solid mass in the liver predominantly on the right lobe. The mass exhibited long T1 T2 signal with restricted diffusion and heterogeneous enhancement after contrast administration (*figure 1*). Cerebral MRI showed no evidence to suggest residual tumor or local recurrence.

Biopsy was proposed, but patient refused and underwent right hemihepatectomy on January, 2020. She was discharged on the seventh post-operative day after an uneventful recovery. Pathology revealed an atypical spindle cell tumor. Considering the previous patient history and previous diagnosis, metastatic meningioma was suggested. The performed immunohistochemical stains showed PR+, EMA+, SSTR2 focally +, Claudin1-. The recurrentbrain tumor showed PR+, EMA+, SSTR2-, Claudin1- (*figure 2*).

With a 12-month follow-up, the patient remains with no evidence of disease recurrence.

We have performed a literature review of 11 previously published liver metastatic cases of meningioma (*table 1*), and associated the present case for analysis.

According to the World Health Organization (WHO)

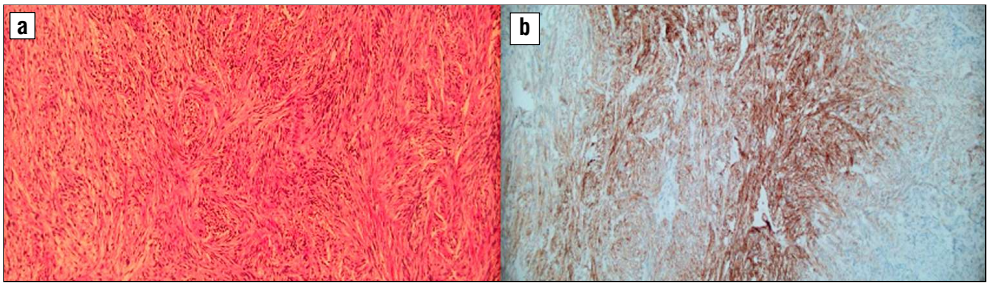

Figure 2 - (a) HE 10 x, HE sections of the liver tumor. The tumor imparts interlacing fascicles of spindle cells with fibrous matrix. (b) EMA 10x, the tumor cells are positive for EMA

Table 1 - Published cases of liver metastasis from meningioma

Case	Sex/age at meningioma presentation	Management of primary	WHO grade of primary	Intra-cranial recurrence	Time from primary to liver met (Months)	Symptoms of liver metastasis	Management of liver metastasis	Status	Follow up time
Present	28, F	Resection	1	Yes, 96 mo.	228	Yes	Resection	Alive, no recurrence	12 months
Ref. (1)	65, F	Resection	2	Yes, 6 mo.	14	Unrelated	Chemotherapy	Deceased	5 months
Ref. (2)	46, F	Resection	Not specified	No	372	Asymptomatic	Observation	Alive, stable with recurrence	5 years
Ref. (4)	65, F	Resection	1	Yes, 180 mo.	Sync with recurrence	Unrelated	Resection	Alive, no recurrence	14 months
Ref. (4)	66, M	Resection	1	Yes, 36 mo.	24	Unrelated	Not specified	Deceased	Not specified
Ref. (6)	54 F	Resection	1	No	Sync with primary	Asymptomatic	Resection	Alive, no recurrence	6 years
Ref. (7)	30, F	Resection	1	Yes, 180 mo.	360	Unrelated	Not specified	Deceased	4 months
Ref. (9)	65, M	Resection	2	Yes, 3 mo.	63	Not specified	Not specified	Deceased	2 months
Ref. (9)	49, M	Resection and radiotherapy	2	Yes, 24 mo.	84	Not specified	Not specified	Deceased	3 weeks
Ref. (12)	61, F	Resection	2	Yes, 24 mo.	84	Yes	Resection	Alive, no recurrence	14 days
Ref. (13)	73, F	Resection	2	Yes, 12 mo.	17	Yes	Radiotherapy	Deceased	Not specified
Ref. (14)	70, MResection and radiotherapy		2	Yes, 48 mo.	Sync with recurrence	Unrelated	Not specified	Not specified	Not specified

classification (5), five of the total 12 cases, (42%) were grade 1, with two (including present case) subsequently recurring intracranially to WHO grade 2.

Ten cases (83%) had 1 or more episodes of intracranial recurrence. The time from first cranial recurrence to the diagnosis of liver metastasis ranged from 0 to 180 months (average 59.5 months). In two patients, liver metastases were synchronous with the primary tumor, and in one, preceded the intracranial recurrence diagnosis by 12 months.

Six out of 12 cases (50%), were either asymptomatic orpresented symptoms unrelated to the liver lesions.

Five patients (42%), were alive, 4 of them with no recurrence after liver resection (14 days-6 years) and one with stable disease (5 years). Six patients were deceased, 2 from the cranial lesions, 1 from the liver lesion, and, in 3, the cause of death was not specified. Vital status was not mentioned in one patient.

DISCUSSION

The most important predictive factor for recurrence and metastasis in meningioma is the tumor grade, according to the WHO criteria (6). Although the rate of malignant change in meningioma is reported to be under 7% (7), metastasis may also occur in grade 1 meningioma without transformation. In fact, any histologically benign meningioma has the potential to metastasize (6), although this tends to occur following one or more episodes of resection forlocal recurrence. In Cecilia L. Dalle Ore et al. research, 100% of metastases were identified in patients with multiply recurrent CNS disease (8). Additional predictive factors include mitotic rate, nuclear atypia, presence of necrosis, and invasionof adjacent vascular structures.

Distant metastases are, nevertheless, rare in meningioma and may easily be overlooked in clinical practice, as most patients remain asymptomatic.

There is no consensus in screening for metastasis after primary meningioma resection. Surov et al. reported that only 50.4% of patients in the literature were symptomatic at the time of diagnosis (10). Cecilia L. Dalle Ore et al. suggest that screening for metastases within the asymptomatic recurrent meningioma population can identify metastases in over 20% of patients (8). Therefore, in our opinion, intracranial meningioma recurrence should constitute a criterion for screening, independently of WHO grade and symptoms. Abdominal ultrasonography, considering its sensitivity, convenience, low cost and non-invasiveness, should be indicated after the first intracranial meningioma recurrence. In the event of any discovered focal liver lesion, 111indium (111In)-octreotide scintigraphy should be applied. Given the higher frequency and density of somatostatin receptors(SSTR) of meningioma, it has the potential to detect extracranial metastases (11) and to differentiate them from other etiologies.

No definite therapeutic regimen has been established for metastatic liver meningiomas. Resection seems to improve the prognosis in metastatic disease, especially if the primary tumor is a low-grade

meningioma. Disease progression or death seem to occur more often in grade II/ III metastatic meningiomas. External beam radiation with its newest components such as photon-based stereotactic radiosurgery can be used for non-resectable lesions. Chemotherapy remains of limited or no proven benefit (6).

CONCLUSION

The majority of primary meningiomas are benign and liver metastasis are rare and often asymptomatic, therefore easilyoverlooked unless actively sought for. We propose that patients with intracranial recurrence of meningioma, even in the benign WHO grade 1, should undergo screening with annual abdominal ultrasound, followed by (111In)-octreotide scintigraphy if any lesion is found.

Disclosures

The authors have no conflict of interest.

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