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James I. Ausman, MD, PhD University of California, Los Angeles, CA, USA

# Case Report

# Paraneoplastic symptoms caused by extracranial meningioma metastases?

# Thomas Mindermann

Neurosurgery, Klinik Im Park, Seestrasse 220, 8027 Zurich, Switzerland

E-mail: Thomas Mindermann - \*tmindermann@hin.ch \*Corresponding author

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## Abstract

**Background:** There are only few reports on distant metastases of cranial meningiomas WHO I. In one-third of the cases, distant metastases seem to be clinically silent. This is the first case of distant metastases which may have manifested with a paraneoplastic syndrome.

**Case Description:** A 52-year-old white male patient was diagnosed with distant metastases to the bones and liver 11 and 12 years following craniotomy and removal of a tentorial meningioma WHO I. At that time, the patient had developed paresthesia, unsteady gait, and a slight cognitive impairment, which in retrospect had no other explanation than that of a paraneoplastic syndrome. Eighteen years following craniotomy, a small intracranial tumor rest is under control following two single session radiosurgery treatments. At present, the patient has a multitude of bone and liver metastases, which seem to cause his paraneoplastic symptoms.

**Conclusion:** Screening for malignancies in patients with paraneoplastic symptoms and a history of cranial meningioma should include screening for distant metastases from the meningioma.



Key Words: Extracranial, meningioma, metastasis, paraneoplastic

# **INTRODUCTION**

The literature on cranial meningiomas WHO I with distant metastases is rare, with many publications including meningiomas WHO II and III.<sup>[1,3-8,10,11,14-17]</sup> The incidence of extracranially metastasizing meningiomas seems to be less than 1:1000.<sup>[7]</sup> If distant metastases occur, they usually affect the lungs, bones, spinal canal, liver, or kidneys. Distant metastases may be present at the time of diagnosis or they may occur as late as more than two decades following the initial diagnosis of intracranial meningioma. Little is known regarding the number, size, clinical symptoms, and signs related to distant metastases or any potential pattern which distant metastases may or may not follow. It appears that almost one-third of distant metastases are clinically silent, which probably leads to underreporting.<sup>[14]</sup>

# CASE REPORT

At 52 years of age, a white male patient underwent craniotomy for a meningioma WHO I of the right tentorium. The postoperative course was uneventful. At the age of 61 years and 9 years following craniotomy, the patient developed paresthesias of the four extremities, an unsteady gait, and problems in concentrating. Based on

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these symptoms and a neurological work up, the patient was diagnosed with peripheral small fiber polyneuropathy. At age 63 and 11 years following craniotomy, a magnetic resonance imaging (MRI) of the pelvis showed a multitude of small skeletal lesions suggestive of metastases [Figure 1]. A tumor screening did not reveal any malignancy. The patient then underwent twice a single session CyberKnife radiosurgery (by TM) for local cranial tumor recurrences at 12 and 17 years following craniotomy. At the time of the second radiosurgery treatment, liver metastases of the meningioma were suspected in a computed tomography (CT) scan and was confirmed bioptically [Figure 2]. In addition, a multitude of vertebral metastases were diagnosed by CT scan at that time [Figures 3 and 4]. At the time of the last follow-up 18 years after craniotomy and at 70 years of age, bone metastases of the lumbar spine were monitored by an MRI [Figure 5]. The patient who is currently 70 years of age is asymptomatic from the initial cranial meningioma. The tumor remnants of the posterior fossa were stable at the current follow-up 18 years after craniotomy or 6 and 1 years following the two radiosurgery treatments [Figure 6]. At present, the patient suffers from back pain, paresthesias of the four extremities, an unsteady gait, and problems in concentrating. He has no focal neurological deficits and no radicular pain. So far, the patient did not undergo focal or systemic treatment for any of the distant metastases.

## **DISCUSSION**

The patient developed distant metastases from cranial meningioma in two of the most frequently affected locations, i.e., the bones and liver. Bone metastases may affect long bones, pelvis, skull, or vertebrae. At least pelvis and vertebrae were affected bones in the present case. The pathophysiology leading to distant metastases may be a hematogenous spread originating from tumor invasion into large venous sinuses or from surgical seeding.<sup>[8]</sup> Benign intracranial meningiomas are

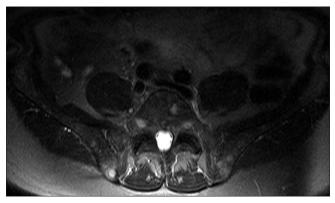


Figure 1: The T2-weighted magnetic resonance imaging of the pelvis shows several bone metastases. The metastases are visible as hypointense lesions within the pelvis bilaterally and the sacrum

slow growing tumors. One of the typical locations of meningiomas are the large venous sinuses such as the superior sagittal sinus or the tentorial sinuses, as in the present case. One may speculate that tumor invasion into the venous sinuses and the resulting contact of tumor cells with the blood flow for over a decade may have led to tumor seeding and distant metastases. Surgical tumor seeding may be another pathway leading to distant metastases. The time for potential seeding is significantly shorter during surgery but the number of cells potentially entering circulation may be much larger. The current patient had both risk factors and eventually he had an enormous number of bone and liver metastases. It is unknown if the tumor had genetic changes which may be another risk factor for aggressive behavior.<sup>[11]</sup> The bone metastases were first diagnosed 11 years following craniotomy and the liver metastases 12 years following craniotomy.

The symptoms of paresthesias, unsteady gait, and problems with concentration began approximately 9 years following craniotomy. One may speculate that those symptoms were paraneoplastic and that the cranial meningioma had metastasized already at that time. A number of paraneoplastic symptoms have been attributed to cranial meningiomas such as optic neuropathy, dermatomyositis, Cushing's syndrome, nephrotic syndrome, or hypocalcemia.<sup>[2,9,12,13,18]</sup> This case may add peripheral small fiber polyneuropathy to the potential list of paraneoplastic symptoms. None of the cases with paraneoplastic symptoms in the literature had distant metastases from the intracranial meningioma. Paraneoplastic symptoms usually subside with the removal of the meningioma. In the present case, a paraneoplastic syndrome developed, even though the intracranial tumor was resected and under control. This was most probably due to the considerable tumor mass of

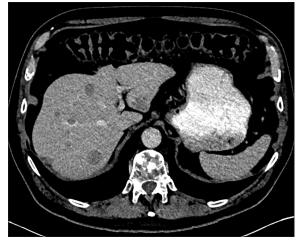


Figure 2:The contrast-enhanced computed tomography scan of the abdomen shows several metastases in the liver 17 years following craniotomy. The metastases are visible as contrast enhancing and hypodense lesions mostly within the right lobe



Figure 3: The sagittal bone window computed tomography scan of the thoracic spine shows several metastases in the vertebrae 17 years following craniotomy. The metastases are visible as hypodense lesions within the vertebrae



Figure 5: The TI-weighted sagittal magnetic resonance imaging of the lumbar spine shows a multitude of bone metastases within the vertebra and the sacrum 18 years following craniotomy. The metastases are visible as contrast enhancing and hypointense lesions within the vertebrae

the numerous liver and bone metastases, which are still in place. There is no proof that the patient's symptoms are paraneoplastic because the tumor mass cannot be removed, which then should lead to resolving symptoms. Yet, it seems to be the most reasonable explanation for the patient's symptoms.

Establishing an accurate diagnosis of metastatic meningioma may be challenging considering that one-third of metastasizing meningiomas are clinically silent and that the disease may manifest with paraneoplastic symptoms, as may have been the case in this patient. At present, there are no recommendations for the treatment of distant metastases other than symptomatic interventions, e.g., in the case of pathological fractures.

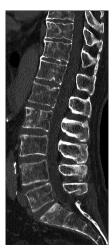


Figure 4: The sagittal bone window computed tomography scan of the lumbar spine shows several metastases in the vertebrae 17 years following craniotomy. The metastases are visible as hypodense lesions within the vertebrae

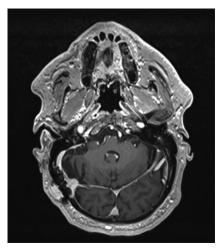


Figure 6: The TI-weighted contrast-enhanced axial magnetic resonance imaging of the head shows a stable tumor rest attached to the right tentorium 18 years following craniotomy. The residual tumor is the small contrast enhancing nodule adjacent to the tentorium and the right tentorial sinus

#### CONCLUSION

This case illustrates that benign intracranial meningiomas WHO I may lead to extracranial metastases and how numerous and yet oligosymptomatic such metastases may be. Patients with distant metastases from cranial meningiomas may present with or without symptoms. They may present with local symptoms and signs caused by the lesions within affected organs or with systemic symptoms caused by paraneoplastic symptoms. To the best of my knowledge, this is the first case of paraneoplastic symptoms, which may be caused by distant metastases from an intracranial meningioma WHO I. Therefore, screening for malignancies in patients with paraneoplastic symptoms and a history of

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cranial meningioma should include screening for distant metastases from the meningioma.

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#### **Conflicts of interest**

There are no conflicts of interest.

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