Benign metastasizing meningioma

Case report

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A patient with a histologically benign intracranial meningioma was found, at the time of initial presentation, also to have a pulmonary tumor. Fine-needle aspiration cytology of the lung mass was consistent with metastatic meningioma. When resected, the pulmonary tumor was found to be histologically benign. The patient has remained well and disease-free for 28 months. Only four other patients with apparently benign metastasizing meningiomas have been described previously.

KEY WORDS • meningioma • metastasis • pulmonary meningioma

WITH few exceptions, the malignancy of a neoplasm is usually defined by its ultimate biological behavior; that is, by its propensity to metastasize. Histological criteria of malignancy are derived from correlations with neoplasms having similar histological features and known malignant behavior. A well known exception to the rule equating metastatic behavior with malignancy is the so-called “benign metastasizing leiomyoma” of uterine origin. Both the primary tumors and pulmonary and lymph-node deposits arising from these uterine tumors are histologically and biologically benign. In rare cases, it appears that certain intracranial meningiomas may exhibit similar behavior.5,10,11

We report a case of a parietal parasagittal meningioma which, at the time of presentation, was found to have spread to the right lung. There were no histopathological features of malignancy in either the primary or secondary tumor, and the meningioma was apparently biologically benign.

Case Report

This 61-year-old non-smoking right-handed man noted the sudden onset of paresthesias in his right leg, right arm, and the right side of his chest while sailing. The symptoms began over about 5 minutes and lasted several hours before subsiding.

Examination. Initial evaluation included frontal and lateral chest roentgenograms which revealed a 2.5-cm rounded area of density in the lower lobe of the right lung (Fig. 1). On the assumption that this mass could represent a primary lung carcinoma presenting...
with intracranial metastases, a computerized tomography (CT) scan of the brain was obtained. Axial and coronal contrast-enhanced CT images revealed a 3-cm left parasagittal mass, which was partially calcified and closely adherent to the falx and cranial vault (Fig. 2 left). The CT findings were interpreted as consistent with a meningioma.

Linear whole-lung tomography confirmed the presence of a solitary nodule in the lower lobe of the right lung, with no evidence of calcification within it. No hilar adenopathy was detected. A percutaneous fine-needle aspiration biopsy of the lung lesion was performed without complication. The cytological diagnosis was "consistent with metastatic meningioma." A planned thoracotomy was then cancelled, and a left internal carotid arteriogram was obtained which revealed displacement of the anterior parietal cortical branches but no abnormal vessels or blush. The mass indented the superior sagittal sinus but did not occlude it. A left external carotid arteriogram showed an enlarged posterior branch of the left middle meningeal artery with a faint tumor blush (Fig. 2 right).

Operations. A left parietal craniotomy was performed. The parasagittal tumor, which arose from the dura and extended into the edge of the superior sagittal sinus was removed. Tumor removal included resection of the edge of the sinus which was closed with a continuous suture. Pathological examination confirmed the clinical diagnosis of meningioma.

Postoperatively, the patient did well. He had some right lower extremity weakness which gradually improved, and he was discharged 2 weeks after surgery. He was readmitted 1 month later, when a right thoracotomy with wedge resection of the right lower lobe nodule was performed. The nodule was histologically identical to the intracranial tumor, and a diagnosis of "metastatic meningioma" was made. The patient is alive and well without any evidence of disease 24 months after his thoracotomy and 28 months since his initial presentation.

Pathological Examination. Specimens from the percutaneous needle aspiration biopsy were prepared both as smears and as filtered fluids, and were stained using the Papanicolaou method. Both types of preparation contained clumps of round-to-oval cells with large pale nuclei; there were numerous clumps in the filtered preparations. The nuclei of the oval cells had a finely stippled chromatin pattern with one to two nucleoli. Many of the clumps showed varying degrees of whorling, including a few well formed tight cellular whorls (Fig. 3 left). Some psammoma bodies were also present.

The 3.5 × 3.0 × 2.7-cm intracranial mass was composed of firm yellow nodular tissue, which was attached to a strip of dura. A 1.3 × 0.4 × 0.3-cm specimen of tumor and dura from the edge of the sagittal sinus was submitted separately. The cut surface of the tumor was white. Hematoxylin and eosin-stained 4-μm paraffin sections, prepared in the usual manner, revealed the tumor to be closely adherent to the dura. Psammoma bodies were very numerous (Fig. 3 center), and the tumor cells formed whorls and lobules with large amounts of interstitial collagen. Some spindle cells were present. Mitoses could not be found despite a careful search of 15 separate sections. There was no necrosis, and no invasion of brain or dura. There was no histological evidence for extension of the tumor into the superior sagittal sinus.

The resected wedge of lung tissue measured 4.5 × 3.2 × 2.2 cm. It contained a firm fleshy white 2.3 × 1.7 × 1.7-cm nodule, which was sharply demarcated from the surrounding tissue. Histological sections revealed a
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transitional meningioma similar to the intracranial tumor, with whorls, psammoma bodies, and abundant collagen. Mitoses and necrosis were absent (Fig. 3 right).

Discussion

Most meningiomas are benign tumors and do not metastasize. If hemangiopericytomas ("angioblastic meningiomas") are excluded, the list of reported cases of meningioma with extracranial metastasis is rather short. Of the 85 cases summarized in 1982 by Kepes, 22 were hemangiopericytic and four others were "angioblastic," leaving 59 cases of metastatic meningiomas published in the world literature since 1886. Some of the older cases were not clearly shown to be meningiomas, further reducing the actual total.

The most common sites to which meningiomas metastasize, in order of frequency, are lungs, liver, lymph nodes, and bones. Almost all of the primary tumors have had histological features conventionally associated with malignancy; that is, they had some combination of focal necrosis, an increased mitotic rate, atypical mitotic figures, and destructive brain invasion. Kepes found only four cases of metastasis from intracranial or spinal meningiomas with benign histopathological features in both the primary and secondary tumors. Of these four, one had a sarcomatous pattern in a recurrent intracranial mass at the time of the patient’s death, although the hepatic metastasis appeared benign. In the second case, the patient died of massive pulmonary metastases 12 years after the original intracranial tumor was excised. The third patient also died from the primary tumor, which had not been operated on. The last case involved a benign-appearing spinal extradural tumor, which eventually recurred locally after it had given rise to radiographically documented pulmonary nodules. The local recurrence had a malignant appearance, as described by Arnould, et al.

Our patient represents an additional case of a benign metastasizing meningioma with good documentation of the "histological benignancy" of both the primary and secondary tumors. In the previously reported cases, the patients died either from the tumor in the primary site, whether recurrent or original, or, in one case, from the metastases; therefore, the biological benignancy of these tumors can be called into question. However, one patient died from a primary tumor without any surgical intervention and the recurrent tumor in a second case was histologically malignant. Given the current 28-month disease-free follow-up period in our patient, the tumors in this case can probably be regarded as biologically benign. However, Ringsted reported a 12-year interval between occurrence of the primary tumor and death of his patient from metastases, so the follow-up findings in our case are not conclusive in this regard.

Extracranial metastases from meningiomas, including malignant neoplasms, have occurred most often following one or more cranial operations. Similarly, spread via intracranial or cerebrospinal fluid pathways usually follows surgery. Of the 59 patients with nonhemangiopericytic meningioma with metastases reviewed by Kepes, all but 13 had undergone surgery well before the discovery of extracranial spread. The present case, however, reemphasizes that distant spread can occur unassociated with any surgery. It has

![Fig. 3. Photomicrographs of tumor specimens. Left: Biopsy specimen obtained by needle aspiration from the lung mass showing a tight cellular whorl. Papanicolaou, Millipore filter preparation, × 425. Center: Specimen from the intracranial meningioma showing psammoma bodies, cellular whorling, lobulation, and areas of spindle cell differentiation. H & E. × 65. Right: Specimen from the pulmonary metastasis showing similar histological features. H & E. × 65.](image-url)
be suggested that the proximity of a meningioma to a dural sinus may make extracranial metastasis more likely, and this may represent the mechanism of spread in this case.

In rare cases, tumors with the histological characteristics of meningioma are found in the lungs with no evidence of an intracranial or spinal primary site despite careful monitoring. Most of these tumors are very small, and correspond to the so-called “minute pulmonary chemodectomas,” or “chemodectoma-type tumors,” which have been shown to be ultrastructurally indistinguishable from meningiomas. Some, however, are quite large, as big as or bigger than the pulmonary mass in our case. It must therefore be conceded that the case we have described could represent the simultaneous discovery of two independent meningiomas—one intracranial and one pulmonary. However, we consider this unlikely, since the pulmonary tumor we report was much larger than the lesions described by Churg and Warnock or by Kuhn and Askin, so that it is unlikely to be an example of a “pulmonary chemodectoma.” More importantly, the pulmonary tumor was histologically so similar to the intracranial tumor that its occurrence from an independent origin strains probability.

Histopathological criteria for malignancy in meningiomas remain at least partly uncertain. Greater than three mitoses/10 high-power fields (HPF) seems to be the most reliable indicator of malignant behavior, but that measure is also imperfect. In a series of 15 cases of malignant meningioma, including hemangiopericytomas, Thomas, et al., reported that seven had between 10 and 30 mitoses/10 HPF. Of the eight others, four were hemangiopericytomas, one was reclassified as benign, and the other three all had at least 1 mitosis/10 HPF, indicating an increased mitotic rate compared to the usual meningioma. These last three were apparently considered “borderline malignant.” New, et al., in a review of seven cases, also reported that mitotic activity above 3/10 HPF correlated with malignancy. Jellinger and Slowik stated that an increased mitotic rate correlated with greater risk for recurrence, but they did not quantify the mitotic rates in their cases.

Fine-needle aspiration biopsy with cytological examination of the specimen obtained has become an important technique in the diagnosis of solitary lung lesions as well as other masses. This is the second case from our institution in which such a biopsy led to the diagnosis of meningioma in the lung. The other case has been reported previously.

In summary, we have reported the case of a patient with an intracranial meningioma of benign histological appearance who at the time of presentation was found to have a metastasis in the lung. The lung neoplasm was histologically similar to the intracranial tumor. The patient is disease-free 28 months after initial presentation. Two similar cases were found on review of the medical literature: in one the metastases killed the patient 12 years after the initial apparently benign intracranial tumor was found, and in the other both the primary tumor and its progeny were first diagnosed at autopsy.

References


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