

Case of Multiple Pulmonary Metastasis from a Benign Intercranial Meningioma

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Abstract

We present the case of a 59 year old male with multiple pulmonary metastases from a previously resected and irradiated atypical WHO grade-2 intracranial meningioma. Grade-2 meningioma is known to have malignant potential although this is rare and accounts for less than 1% of the total cases of meningioma. The lung is the most common organ of metastases accounting for 61% of documented cases.

Introduction: Meningiomas are considered the most common intracranial tumours. Most are benign, accounting for 94% of meningiomas and rarely metastasise (<1% of meningiomas). They can be divided into three grades, typical/benign (WHO grade-1), atypical (WHO grade-2) and anaplastic (WHO grade-3).

Case description: We present the case of a 59-year-old male with a background of previously excised meningioma followed by radiotherapy, who presented with bilateral metastatic lung lesions 7 years after primary treatment, which was subsequently confirmed to represent metastatic meningioma through histological examination.

Discussion: Multiple pulmonary metastases from intracranial meningiomas are rare. Only 11 cases of pathologically confirmed WHO grade-2 intracranial meningiomas with multiple pulmonary metastases have been documented in the literature. Surgical resection is the treatment of choice for accessible intracranial meningiomas with radiotherapy currently recommended for prevention of local recurrence. WHO grade-2 meningiomas, often require adjuvant treatment with radiotherapy or stereotactic

radiosurgery. New approaches which combine the use of chemotherapy and targeted treatments are currently ongoing in clinical trials.

Conclusion: Long-term outcomes for patients with meningioma pulmonary metastases are difficult to study due to its rarity. However, metastatic meningioma should be included in the differential diagnosis of cannon ball pulmonary lesions where there is a history of meningioma, especially with local intra-cranial recurrence.

Keywords

Tumor; Carcinomas; Anorectal; Cancer; Radiotherapy

Introduction

Mostly benign with slow growth rate, accounting for 94% of cases, and rare tendency to metastasise (<1%) meningiomas are the most common primary intracranial tumours [1]. Broadly divided into three histologic grades, meningioma can be typical/benign (WHO grade-1), atypical (WHO grade-2) or anaplastic (WHO grade-3). Most are grade-1 (90%) with grade-2 and grade 3 histology subtypes representing 5%–7% and 1%–3% of all meningiomas respectively.

Metastasis from atypical and anaplastic meningiomas has been reported in up to 5% and 30% of the cases respectively. Whilst meningiomas are more common in women, the metastatic variants are more commonly encountered in men [2]. Review of the literature shows that the lung is the most common organ for metastasis (61%) followed by the pleura, liver, abdominal viscera, lymph nodes and bones [2].

Here we report a rare case of multiple pulmonary metastases from a WHO grade-2 meningioma.

Case Presentation

A 59 year old Caucasian male presented to our institution in June 2020 following a witnessed seizure. There was a background

history of previously excised WHO grade-2 frontal meningioma and stereotactic radiation therapy, (60 Grey delivered in 30 fractions) in 2013. Other previous history included further debulking surgery in 2018 due to significant growth of almost three times in a relatively short interval period of 6 months with suspicion of focal brain invasion.

A CT brain performed on presentation to our Emergency department, demonstrated no interval change from multiple prior surveillance studies, subsequently characterised by MR brain with contrast as post-craniotomy gliosis and post-radiation necrosis at the site of primary disease.

Surprisingly, a routine chest radiograph performed to investigate anterior chest wall pain (**Figure 1**), demonstrated multiple, well circumscribed non-cavitating round opacities (cannon ball lesions) in the lungs bilaterally measuring up to 60 mm. Importantly, the clinical history was devoid of respiratory symptoms such as cough and dyspnoea.



Figure 1 Multiple cannon ball opacities in the lungs bilaterally measuring up to 60 mm in keeping with metastatic disease.

CT Thorax imaging (**Figure 2**) subsequently revealed multiple pulmonary metastases. Of note the patient last chest imaging was from 2013 which showed no evidence of metastatic disease. No synchronous neoplastic pathologies to account for the metastatic disease in the lungs were identified and further investigation with sigmoidoscopy and endoscopy were reported as normal.



Figure 2 A selective slice on coronal (A) & axial (B) plane CT image depicting multiple pulmonary lesions.

The patient went on to have a bronchoscopy and an Endobronchial Ultrasound-guided Transbronchial Needle Aspiration (EBUS) with the cytology from the specimens suggestive of metastatic meningioma with further management pending review of the options with the patient at his first clinical follow up.

Discussion

Multiple pulmonary metastases from intracranial meningiomas are rare. A recent case series found 11 patients with a pathologically confirmed WHO grade-2 meningioma with distant metastasis to the lung [3]. All patients included in the study had either local tumour recurrence, surgical resections, or had undergone radiotherapy at least once.

The patients with pulmonary metastasis are rarely symptomatic and typically present with single or multiple round, well circumscribed noncalcified parenchymal nodules of varying size on follow up or routine imaging. Multiple deposits are identified in 50% of cases.

The average time from diagnosis of the primary tumour to discovery of metastases is 6 years [4] with the longest documented interval of 24 years [5]. In addition, metastases are frequently discovered following recurrence of the primary lesion.

Our case demonstrates all classic features of metastatic pulmonary disease from treated recurrent meningioma, highlighted in the literature.

Risk factors for metastasis include previous craniotomy, venous sinus invasion, local recurrence, and aggressive histological subtypes. Hematogenous metastasis may be the most likely mechanism for extracranial spread with 70% of patients reporting to have had metastatic disease following a craniotomy [4]. Indeed, Figueroa et al [6] revealed that 75% of patients with metastasizing meningioma gave history of previous surgery on the primary lesion or invasion of the venous sinuses.

A second route believed to lead to the development of distant metastasis may be via the cerebrospinal fluid pathway [7]. It is hypothesised that surgical interference may release the tumour cells from its cohesive state into the cerebrospinal fluid. The local recurrence rates even after complete resection vary from 9 to 32%. Despite relatively large number of surgical resections performed each year, metastasis remains rare at less than 1% [8].

There is no standard treatment with curative intent for metastatic meningioma. Complete surgical resection for accessible intracranial meningiomas is the mainstay of treatment for WHO grade-1 meningiomas, whereas grade-2 and 3 tumours often require adjuvant treatment, either radiotherapy or stereotactic radiosurgery. Postoperative radiotherapy has been recommended for prevention of local recurrence, especially when resection is subtotal or when the histology is suggestive of an aggressive histology [9]. Surgical resection of pulmonary lesions is restricted to those patients who have no extra-thoracic metastatic disease and only if all metastatic lesions could be excised without affecting residual pulmonary function. New approaches which combine the use of chemotherapy and targeted treatments are currently ongoing in clinical trials [10-12].

Differential diagnosis

The differential diagnosis for multiple cannon ball pulmonary metastasis includes renal cell carcinoma, choriocarcinoma, sarcoma, thyroid carcinoma and metastasis from prostate cancer [13]. Benign causes of multiple pulmonary metastasis include uterine leiomyoma and giant cell tumour of the bone in addition to meningioma.

Conclusion

In conclusion, metastasis from meningioma should be considered in patients presenting with multiple cannon ball pulmonary lesions and history of meningioma.

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