

Multiple pulmonary metastases from asymptomatic benign intracranial meningioma: case report

Intrakranijinės besimptomės meningiomos daugybinės metastazės į plaučius: klinikinis atvejis

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Background

This case presents unexpected association between diseases and symptoms. We believe that it should compel clinicians to rethink the known behaviors of the benign tumors, in particular meningioma which can gain independent metastatic potential. This report aims to increase the awareness of clinicians toward patients with this unusual and clinically isolated pattern, because metastases can remain misdiagnosed for a long period of time. This case contributes to medical knowledge, diagnostic and prognostic approaches. This is also the first case of metastatic meningioma reported in Lithuania.

Case presentation

We report a 66-year-old woman who presented with persistent productive cough, dyspnea, fever and weakness during physical activity. Chest radiographs revealed multiple small round shaped pulmonary nodules. Thoracoscopic resection and histopathology showed a benign meningioma. Magnetic resonance imaging scan of the brain demonstrated a small dural based mass in the left pontocerebellar angle connected with deep dilated cerebral venous network and superior petrosal sinus. Morphological findings of the session were consistent with a World Health Organization grade I fibrous meningioma.

Conclusion

Meningiomas are usually non-invasive tumors and do not metastasize and hence, are perceived as benign tumors. Patients with this uncommon and clinically isolated pattern of metastases can remain misdiagnosed for a long period of time, due to unexpected behavior of this particular tumor. This case report denies most of the criteria of possible risk factors for the development of metastases from a meningioma what allows to consider it as tumor with unpredictable behavior.

Key words: case report, meningioma, benign, asymptomatic, metastases.

Įvadas

Šis klinikinis atvejis yra nenumatyto ligos pasireiškimo. Tikime, kad jis privers apgalvoti jau žinomą gerybinių navikų, tarp jų ir meningiomas, elgseną, kuri gali įgyti nepriklausomą metastazinį potencialą. Kadangi gerybinių navikų metastazės yra diagnozuojamos pavėluotai, šiuo pavyzdžiu siekiame plėsti klinicistų sąmoningumą ir budrumą nagrinėjant ligas, kurioms būdingos neįprastos klinikinės apraiškos. Tai pirmasis Lietuvoje aprašytas intrakranijinės meningiomos metastazavimo į plaučius atvejis.

Atvejo pristatymas

Aprašome klinikinį atvejį 66 metų moters, kurią vargino nuolatinis produktyvus kosulys, dusulys, karščiavimas ir silpnumas, atsirandantis fizinio krūvio metu. Atliktos krūtinės ląstos rentgenogramos parodė daugybinius, mažus, apvalios formos mazgus plaučiuose. Po atliktos torakoskopinės plaučių audinių rezekcijos histopatologinio tyrimo metodu konstatuota meningioma. Atliktas galvos smegenų magnetinio rezonanso tyrimas, kurio išvada – kairiajame tilto ir smegenėlių kampe su dangalais susijęs, kontrastinę medžiagą gerai kaupiantis, smulkus navikas, išsiplėtusios giliosios smegenų venos. Histologinė smegenų darinio išvada remiantis Pasaulio sveikatos organizacijos klasifikacija – pirmo laipsnio fibrozinė meningioma.

Išvada

Meningioma yra traktuojama kaip neinvazinis ir nemetastazuojantis, gerybinis, intrakranijinis navikas. Metastaziniai židiniai yra neįprasti šios ligos eigai, dėl to gali būti diagnozuoti pavėluotai. Klinikinis atvejis paneigia literatūroje nurodytus rizikos veiksnius, kuriais remiantis būtų galima numatyti galimą meningiomos metastazavimą, ir tai leidžia priskirti meningiomą neprognozuojamos eigos navikams.

Reikšminiai žodžiai: klinikinis atvejis, meningioma, gerybinis navikas, besimptomė eiga, metastazės

Background

Meningioma is the most common primary extra-axial tumor of the central nervous system in adults [1, 2], and arises from the meningoepithelial cells of the leptomeninges. Extracranial metastases of meningiomas are rare and occur in less than 1 per 1000 cases [3, 4, 5, 6]. The most common site of metastasis is the lung, which counts for 61% of all meningioma metastases [7, 8], followed by the liver, lymph nodes, and bones [9]. There are only isolated case reports, regarding pulmonary meningioma metastases, most of which, as the presented case, were incidentally detected by chest radiography or Computed tomography (CT) scans [10, 11].

We report a case of 66-year-old woman who presented acute symptoms of pneumonia. Chest X-ray revealed pulmonary metastases, the origin of which turned to be asymptomatic benign meningioma of pontocerebellar region.

Case presentation

A 66-year-old woman presented with persistent productive cough, dyspnea, fever and weakness during physical activity. Respiratory system examination displayed stable hemodynamic, bronchovesicular breathing on both sides, persistent cough with observable watery phlegm was noted. Laboratory testing showed increased white cells count. Firstly pneumonia was expected, but chest radiographs unveiled multiple small round shaped pulmonary nodules (Fig. 1A). This discovery led to the necessary histopathology testing to discard other possible diseases forming nodules such as tuberculosis or malignant tumors. The patient was referred to a local university hospital for further analysis.

Diagnostic thoracoscopic resection of mediastinal masses was performed and histopathology showed meningioma (World Health Organization (WHO) grade I) (Fig. 2A). Lung mass was positive for Epithe-

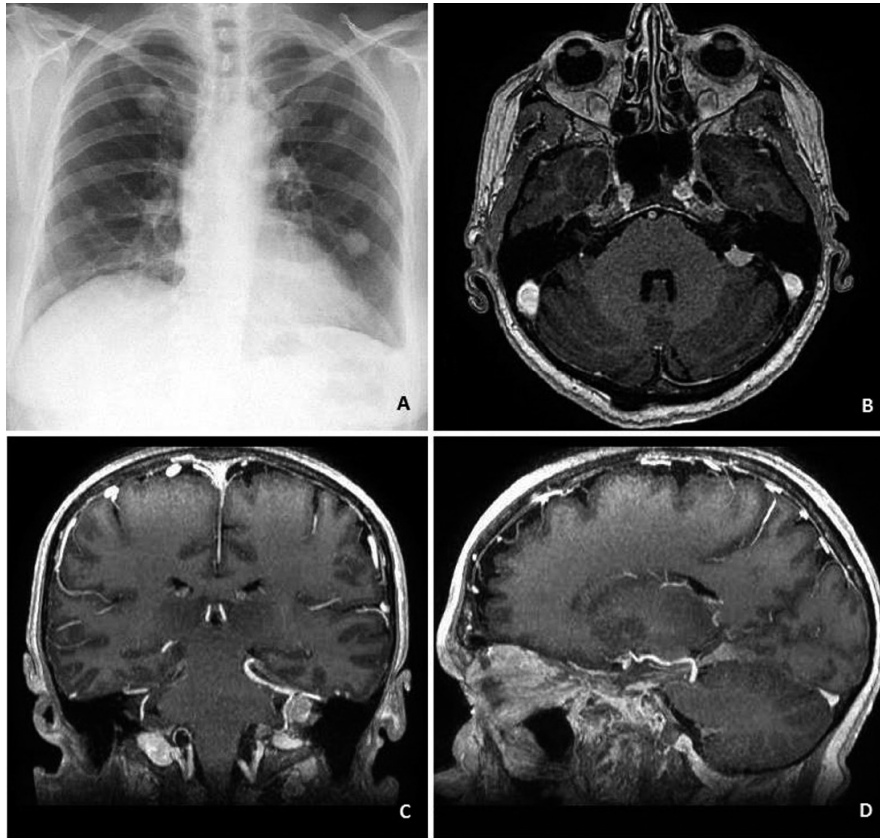


Fig. 1. Radiological findings
 A. Chest radiograph shows multiple round shaped lesions in both lobes.
 B. C. D. Contrast enhanced T1 brain MRI shows the dural-based homogeneously enhancing mass (9 × 6 mm) at left ponto-cerebellar angle with dilated deep cerebral veins and superior petrosal sinus

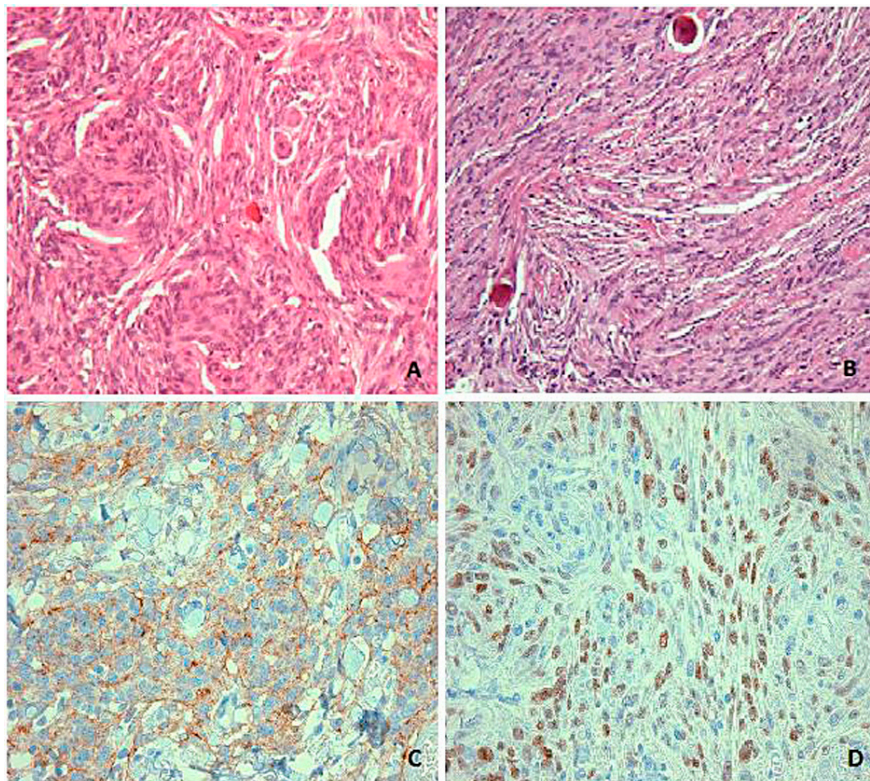


Fig. 2. Morphological findings in consistent with a WHO grade I fibrous meningioma
 A. Histopathological examination of the pulmonary lesion shows oblong-shaped cells with oval nuclei and intranuclear pseudoinclusions.
 B. Histopathological examination of brain mass shows oblong-shaped cells with slightly polymorphic nuclei, abundant eosinophilic cytoplasm and average cellularity, without mitotic activity. The tumor consists of meningotheial cells arranged in fascicles with psammoma bodies.
 C. Lung mass positive for EMA (cytoplasmic staining)
 D. Lung mass positive for PR (nuclear staining)

Risk factors for the development of metastases from a meningioma include histological criteria such as:

1. cellularity,
2. cellular heterogeneity,
3. high mitotic rate,
4. nuclear pleomorphism,
5. tumor necrosis,
6. invasion of adjacent blood vessels.

Case report findings
<ul style="list-style-type: none"> • oblong-shaped cells • slightly atypical nuclei • average cellularity, without mitotic activity • cells arranged in fascicles • very low Ki-67 proliferation index

Kanthan R, Senger JL. Distant metastases from meningiomas – A myth or reality? 2013; *Ann Clin Pathol* 1(1): 1001.

The literature suggests that

- craniotomy,
- venous sinus invasion,
- local recurrence,
- papillary morphology,
- histological malignancy

Case report findings
<ul style="list-style-type: none"> • no previous craniotomy • no venous sinus invasion • no local recurrence • no papillary morphology • grade I fibrous meningioma

may be risk factors for systemic spread.

T. Eran, I. Hanta, S. Hacıyakupoğlu, S. Zorludemir, H. Zeren, and A. I. Göçer. Huge bilateral pulmonary and pleural metastasis from intracranial meningioma: A case report and review of the literature. *Journal of Neuro-Oncology*, vol. 74, no. 2, pp. 179-181, 2005.

Adlakha A, Rao K., Adlakha H, Perry A, Crotty T, Scheithauer BS et al. Meningioma metastatic to the lung. *Mayo Clinic Proceedings*, vol. 74, no. 11, pp. 1129-1133, 1999.

lial membrane antigen (EMA) (cytoplasmic staining (Fig. 2C)) and Progesterone receptor (PR) (nuclear staining (Fig. 2D)). Immunohistochemical stainings for CD34, CD117, CD99, Actin, Desmin, S100, panCK, TLE-1 were negative.

Patient was conscious and oriented, neurological examination showed no signs of neurologic deficit: symmetrical pupils with positive photoreaction and free eye movement; symmetrical face with sensations not affected, and no visible meningeal symptoms. Limb tone, movements and tendon reflexes without any pathological symptoms. Magnetic resonance imaging (MRI) scan of the brain revealed the dural based contrast enhancing mass (9x6 mm) at the left ponto-cerebellar angle connected with dilated venous network, deep left cerebral veins and superior petrosal sinus (Fig. 1B). No perifocal reaction or mass effect was found.

Total surgical resection of the tumor mass via left retrosigmoidal approach was performed. Histopathological examination of the brain mass (Fig. 2B) showed oblong-shaped cells with slightly atypical nuclei, abundant eosinophilic cytoplasm and average cellularity, without mitotic activity. The tumor consists of meningotheial cells arranged in fascicles with psammoma bodies. Ki-67 proliferation index was very low. Morphological findings were consistent with a WHO grade I fibrous meningioma.

Histopathological examination parameters of the intracranial tumor matched those of the lung mass. Histological findings together with the clinical history were

suggestive of a metastatic meningioma WHO grade I.

After the surgery, patient showed no postoperative neurological deficiencies. Within first twenty-four hours, patient was treated with analgetics, single dose of antibiotics (Cephazoline) and anticoagulant for deep vein thrombosis prophylactics. Physical rehabilitation was started for early vertigo control. After surgery CT showed small air spaces and postoperative alteration with no signs of bleeding. Surgical incision healed without any contamination, sutures were removed and patient was discharged from hospital. It was recommended to consult a neurologist and neurosurgeon after 6 months for examination and control MRI checkup.

After half a year, MRI examination showed no signs of brain tumor recurrence. Currently the patient is being treated for lung masses in her local hospital. In view of the slow-growing nature of these metastases and their good prognosis after resection, surgery is the treatment of choice [12].

Discussion

Meningiomas (WHO grade I) are usually non-invasive tumors and do not metastasize and hence, are perceived as benign tumors [12]. However, a larger number of meningiomas (approximately 20%) exhibit aggressive behavior, with approximately 1% of all patients with meningiomas developing distant metastases [13, 14, 15, 16].

Risk factors for the development of metastases from a meningioma include histological criteria such as cel-

lularity, cellular heterogeneity, high mitotic rate, nuclear pleomorphism, tumor necrosis, and invasion of adjacent blood vessels [17]. In our patient, review of the histological appearances of the primary and metastatic tumor revealed no signs of malignancy. Case showed that, a high cell proliferation rate and other histological criteria are not essential for the occurrence of extracranial metastases and histologically benign meningioma can possibly metastasise [5].

The literature suggests that previous craniotomy, venous sinus invasion, local recurrence, papillary morphology, and histological malignancy may be risk factors for systemic spread [18, 19]. As seen in the case of our patient, metastatic meningioma may present

without previous local tumor recurrence or histological malignancy.

Conclusions

The present case report emphasize the importance of performing careful examination for patients, to detect and treat any distant metastases as early as possible, even though they occur rarely. The benign meningioma not only can become malignant, but also can acquire independent metastatic potential and metastasize outside central nervous system. In reference to our case, we can state that there are no definitive criteria to predict local recurrence or metastases what allows to consider meningioma as tumor with unpredictable behavior.

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LIST OF ABBREVIATIONS

CT – Computed tomography

EMA – Epithelial membrane antigen

MRI – Magnetic resonance imaging

PR – Progesterone receptor

WHO – World Health Organization