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Metastases of intracranial meningioma with sarcomatous change to the lung: a case report

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Metastases of meningioma outside the central nervous system are extremely rare (0.1% of all cases). We report a case of recurrent sarcomatous meningioma with the metastatic spread to the lung. A 34-year-old man had a left parietal tumour which was subtotally removed and diagnosed as a transitional meningioma. He received a post-operative radiotherapy of 60.6 Gy. The local recurrence of identical histological appearance was subtotally resected eight years later. Two years after the second craniotomy two of the three revealed pulmonary tumours were resected and diagnosed as a sarcomatous meningioma. Simultaneously disclosed second local recurrence was re-irradiated to dose of 30 Gy. Patient is alive with stable neurologic performance status at 6 months after the radiation retreatment. We also discuss the factors associated with extracranial metastases in meningioma.

Wewnątrzczaszkowy oponiak z komponentem mięsakowym z przerzutami do płuc – opis przypadku

Przerzuty oponiaków poza ośrodkowy układ nerwowy występują niezwykle rzadko (0,1% przypadków). W artykule opisano przypadek nawrotowego oponiaka wewnątrzczaszkowego z przerzutami do płuc. U 34-letniego mężczyzny z miesięcznym wywiadem napadów padaczkowych oraz narastającego niedowładu połowiczego stwierdzono w badaniu tomografii komputerowej obecność guza sklepistości lewej półkuli mózgu. Przeprowadzono subtotalną resekcję i stwierdzono oponiaka typu przejściowego – meningioma transitionale. Chorego napromieniono pooperacyjnie, podając dawkę 60,6 Gy w 36 frakcjach na obszar guza pierwotnego z 2 cm marginesem. Po 8 latach obserwacji wystąpiła wznowa miejscowa, którą poddano częściowej resekcji, potwierdzając wcześniejsze rozpoznanie. Dwa lata później ujawniono 3 zmiany przerzutowe w płucach. Dwie z nich usunięto, stwierdzając oponiaka z komponentem mięsakowym. Jednocześnie wykonane badanie rezonansu magnetycznego mózgu wykazało dalszą miejscową progresję. Chorego ponownie napromieniono, podając dawkę 30 Gy w 10 frakcjach wyłącznie na obszar guza. Chory żyje 9 miesięcy po usunięciu zmian przerzutowych i 6 miesięcy po powtórnej radioterapii wznowy. Jego stan neurologiczny i radiologiczny obraz guza nie uległy zmianie.
Dokonano przeglądu piśmiennictwa dotyczącego podobnych przypadków, zwracając uwagę na czynniki zwiększające ryzyko powstania przerzutów pozaczaszkowych oponiaków mózgu.

Key words: meningioma, extracranial metastases, recurrent meningioma
Słowa kluczowe: oponiak, przerzuty pozaczaszkowe, nawrotowy oponiak

Introduction

Extracranial metastases in meningioma occur in only 0.1% of all cases [1, 2]. The lung is the most common site (over 60 %) [1, 3, 4]. Other sites, in order of frequency, are liver, bone, pleura, mediastinum, lymph nodes, kidneys [1, 3-5]. Histologically, the metastases are usually similar to the primary lesion [3]. Most patients have single metastasis but multiple metastases are not rare. We report a case of pulmonary metastases from recurrent intracranial meningioma with sarcomatous change.

Case report

A 34-year-old man presented with a 1-month history of seizures and a mild right hemiparesis. CT scans showed in the left parietal lobe a 4.5 cm tumour which extended from the falx to the convexity of the left cerebral hemisphere and was surrounded by oedema zone. Subtotal resection of the tumour was performed and a diagnosis of transitional meningioma was established by pathological examination. The operation was followed by radiation therapy. Patient received a dose of 60.6 Gy in 36 fractions to the preoperative tumour area with a margin of 2 cm.

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There were rare seizures after the treatment and therefore carbamazepine was administered. At 8 years of follow-up period the patient developed strong seizures and imaging procedures demonstrated local tumour recurrence. A partial resection was performed and the pathological examination was interpreted as a transitional meningioma. A chest radiography was without abnormalities.

Almost 2 years later, imaging studies disclosed a 3.5 cm tumour in the left upper lung lobe and two tumours of diameter 1.0 and 1.5 cm in the right lower lung lobe. Two of them were found at surgery and resected. The histological diagnosis was metastatic transitional meningioma. The review of the specimen by the consulting pathologists revealed features of malignancy. Finally the diagnosis was: sarcomatous meningioma. Simultaneously made MRI of the brain disclosed multiple meningiomas of the falx and convexity of the right parietal lobe. The patient received radiation dose of 30 Gy to this site. The patient is alive with stable pulmonary disease 9 months after thoracic surgery and the cerebral disease remains unchanged 6 months after second course of radiation therapy. The moderate right hemiparesis persists without any improvement from the presentation of the disease. No toxicity of the twice delivered radiotherapy was observed at the follow-up period.

Discussion

The resectability of the tumour corresponds closely to the clinical outcome in meningiomas [6, 7]. According to the published data, 5- and 10-years survival in benign meningiomas treated by surgery and radiation were respectively 80 and 70% [7]. This implies the long period to the development of metastases. Stoller et al. [8] reported that the mean interval from detection of the primary tumour to detection of the first metastasis was 77 months (6.4 years) +/- 66 months. In presented case this interval was almost 8 years long.

Meningiomas metastasize extremely rarely (0.1% of all cases is reported on review of the medical literature) [1, 2]. Among these data probably about one third were metastasising haemangiopericytomas, which according to the recent World Health Organisation classification form distinct pathological entity [1, 5].

In relation to the described case we would like to analyse some features which are mentioned as factors predicting metastases in meningiomas. They are:

- histopathological features [1], especially papillary variant with a significantly higher rate of metastatic spread than in meningiomas as a whole,
- surgery management of primary and recurrent tumour [1, 6],
- venous sinus invasion [8],
- neuroimaging signs with peritumoural oedema, heterogeneous contrast enhancement, minimal or no calcification, indistinct or irregular margins [9].

According to the published data, histologic pattern does not always relate to the development of metastases [6]. Although most metastasising meningiomas have features of malignancy [1, 2] there are also reported cases of metastasising benign meningiomas [3, 6, 10]. On the other hand, with recurrences, benign meningiomas can develop malignant transformation, while the metastases themselves are also malignant or are free of morphologic features of malignancy [6, 11-13]. In presented case, recurrent tumour probably underwent malignant transformation after an initial treatment, but the part of tumour resected during the second craniotomy was without any features of malignancy. It may be related to the heterogeneity of brain tumours and the probability of resection or examination of only benign part of the tumour. The metastases to the lung, disclosed two years later, related to the intracranial recurrence, were malignant.

Although distant spread can occur in the absence of previous surgery [3, 5] many authors claim that extracranial metastases have occurred most often following one or more craniotomies [6, 8, 10, 13]. Figueroa et al. [1] estimate that metastases are associated with prior surgery or the invasion of a venous sinus in 75% of cases. In the presented report the invasion of the superior sagittal sinus was confirmed and the metastases were revealed 2 years after the second craniotomy.

Some authors suggest an invasion of venous sinus as a risk factor related to occurrence of hematogenous extracranial metastases [8]. According to others the metastases seem to be related neither to the location nor to the size of the intracranial primary [1, 4].

Radiographic patterns regarded as potential of malignancy seem to be useful to predict more aggressive behaviour in these cases in which partial resection or biopsy of intracranial mass provide only a specimen from benign part of tumour.

The next frequently asked question is the way of metastasising. Kruse et al. [10] stated that metastases to the liver and kidneys are blood-borne from pulmonary tumours. He suggested that in cases in which pulmonary metastases were not noted they could be overlooked or had undergone regression. Our patient have had only the pulmonary metastases. Therefore the careful monitoring of other organs is desirable.

Conclusions

Presented case of meningioma fulfilled at least four conditions when aggressive behaviour and distant spread are more probable than in other intracranial meningiomas. These are recurrence, repeated craniotomies, venous sinus invasion and probable malignant transformation.

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