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Multiple Meningiomas with Different Histological Patterns in the Same Patient: Do They Exist? A Case Report and Literature Review

Saranraj M.K.[1] Salapathi Shanmugam[2] Yatin Raiyani[1] Adrian Jacob[1] Siddhartha Ghosh[1]

[1] Neurosurgery Department, Apollo Hospitals, Vanagaram, Chennai, Tamil Nadu, India
[2] Pathology Department, Apollo Hospitals, Vanagaram, Chennai, Tamil Nadu, India

Address for correspondence Saranraj M.K., DrNB Resident, Neurosurgery Department, Apollo Hospitals, Vanagaram, Chennai, Tamil Nadu, India (e-mail: mks0434@gmail.com).

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Abstract

Meningiomas are the most common form of primary intracranial tumors, accounting for 13 to 26% of total neoplasms arising from arachnoid cap cells of the meningeal layer covering the central nervous system. Multiple intracranial meningiomas, which often have a syndromic association, account for less than 10% of total meningiomas. Multiple meningiomas with different histological patterns or grades in the same patient are very rare. Here we report such a rare case of meningioma with different histological patterns in the same patient. A 56-year-old lady presented to us with complaints of progressive right-sided weakness and speech disturbances, and her magnetic resonance imaging (MRI) showed two distinct extra-axial lesions over left frontal convexity and left fronto-temporo-parietal convexity. She underwent left fronto-temporo-parietal craniectomy and Simpson grade 1 excision of the lesions. Her histopathological examination revealed two different histological patterns: lesion 1 (left fronto-temporo-parietal convexity) was reported as a transitional World Health Organization (WHO) grade 1 meningioma and lesion 2 (left frontal convexity) was reported as angiomatous WHO grade 1 meningioma. She recovered well and was discharged in a stable condition after 3 weeks. Multiple meningiomas are defined as at least two spatially separated meningiomas occurring at the same time or more than two meningiomas arising sequentially from two clearly distinct regions. The exact mechanism of multicentricity is unknown. The treatment protocol for different histological types in the same patient is not clear. The extent of surgical resection remains the mainstay of the treatment and these patients should be followed up closely to watch for recurrence or malignant transformation. The role of radiotherapy in multiple meningiomas is yet to be established.

Keywords

- dual pathology in multiple meningioma
- intratumoral variation
- multiple meningioma
- Simpson 1 excision
- transitional and angiomatous meningioma

Introduction

Meningiomas are the most common form of primary intracranial tumors, accounting for 13 to 26% of total neoplasms arising from arachnoid cap cells of the meningeal layer covering the central nervous system.[1,2] The current World Health Organization (WHO) grading system (2021) classifies meningiomas based on their histopathological appearance into grades 1 to 3. Various subtypes were described under all grades based on the histological patterns. Multiple intracranial

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meningiomas accounts for less than 10% of total meningiomas, which often have a syndromic association commonly neurofibromatosis 2, where sporadic occurrence is very uncommon.[1,2] Cases of multiple meningiomas (MMs) with different histological patterns or grades in the same patient are very rare.[2] To the best of our knowledge, only 20 cases have been reported in the literature so far.[2–6] Here we report such a rare case of meningioma with different histological patterns in the same patient.

Case Report

A 56-year-old lady presented to us with a history of gradually progressive right-sided weakness of both upper and lower limbs with facial deviation to the left for 1 year, altered sensorium, and speech disturbances for 6 months. On examination, she was conscious, dysphasic with right hemiparesis and right UMN facial paresis. No neurocutaneous markers were noted. Her magnetic resonance imaging (MRI) of the brain with contrast showed a large extra-axial iso- to hypointense on T1-weighted imaging (T1WI) and mild hyperintense lesion on T2WI over the left fronto-temporo-parietal region with a broad base over the convexity meninges. It also showed a homogenous moderate enhancement on contrast administration with multiple flow voids inside the lesion. Another lobulated extra-axial hypointense on T1WI and extremely hyperintense lesion on T2WI was observed over the left frontal convexity, and on contrast administration, the lesion showed intense contrast enhancement (►**Fig. 1**). Perilesional edema was seen around both the lesions. MR angiogram showed enlargement of the left superficial temporal artery and middle meningeal artery. All the features were consistent with a meningioma. She underwent left fronto-temporo-parietal craniectomy and Simpson grade 1 excision of the lesions. The postoperative period was uneventful. Her histopathological examination revealed two different histological patterns: lesion 1 (left fronto-temporo-parietal convexity) was reported as a transitional WHO grade 1 meningioma and lesion 2 (left frontal convexity) was reported as an angiomatous WHO grade 1 meningioma (►**Fig. 2**). She recovered well and was discharged in a stable condition after 3 weeks.

Discussion

Meningiomas are the most common nonglial primary intracranial neoplasm, accounting for 13 to 25% of the total intracranial neoplasms arising from arachnoid cap cells of the meningeal layer.[1,2] MMs are characterized by the occurrence of at least two meningiomas that are spatially separated at the same time or by the sequential emergence of more than two meningiomas from two obviously distinct locations.[1,5,6] MMs are frequently found in patients with neurofibromatosis type 2 (NF2); however, they occur much less frequently in cases of sporadic meningioma.[2] MMs account for less than 10% of the total cases.[1,2] MMs with different histological patterns or grades in the same patient are very rare. A total of 20 patients have been reported to date with different histological patterns in the same patient.[1–8] A combination of concurrent fibrous and atypical

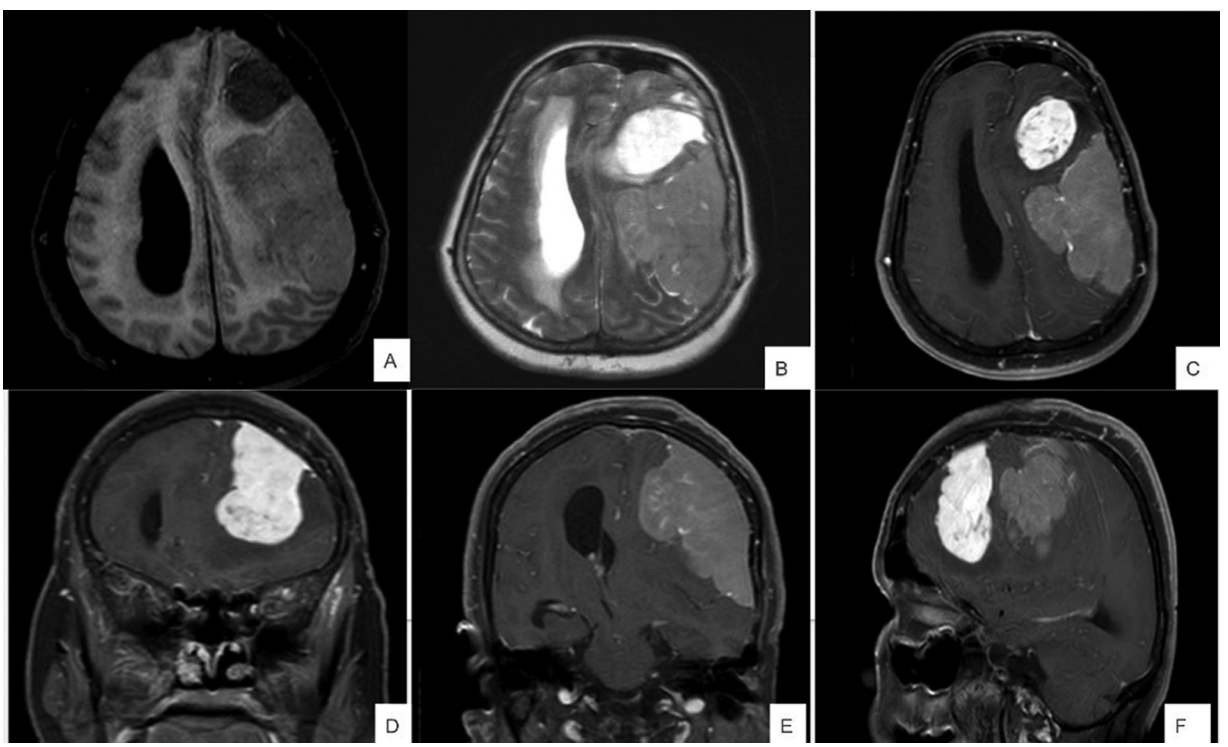

Fig. 1 (**A**) A large extra-axial iso- to hypointense on T1-weighted imaging (T1WI) and (**B**) Hyperintense lesion on T2WI over the left fronto-temporo-parietal region with a broad base over the convexity meninges and (**C, E, F**) homogenous moderate enhancement on contrast administration with multiple flow voids inside the lesion. Another lobulated extra-axial (**A**) hypointense lesion on T1WI and (**B**) extremely hyperintense lesion on T2WI over the left frontal convexity. (**C, D, F**) On contrast administration, the lesion showed intense contrast enhancement.

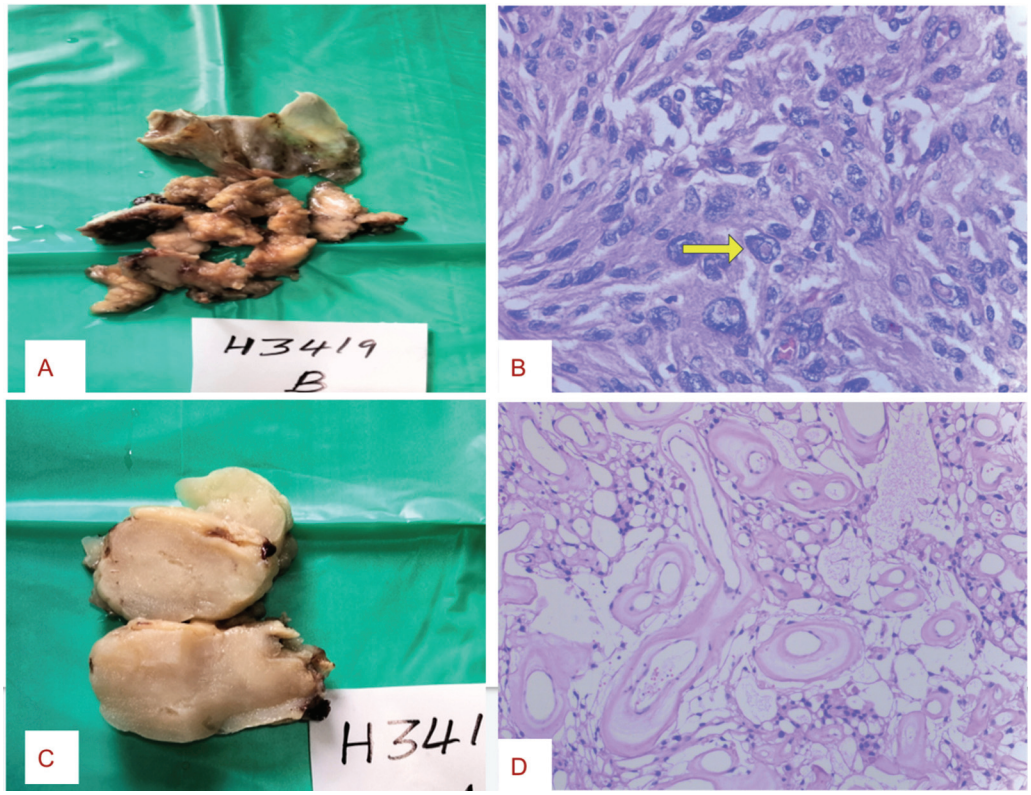

Fig. 2 (A) Multiple fragments of grayish white firm tissue. (B) A section from lesion 1 shows oval cells in fascicles and intranuclear inclusion bodies (*arrow*), hematoxylin and eosin (H&E), 40X. (C) Grayish white homogenous tissue with spongy appearance. (D) A section from lesion 2 shows tumor cells with interspersed varying sizes of hyalinized vessels (H&E, 40X).

meningioma, psammomatous and atypical meningiomas, fibrous meningioma, transitional meningioma, meningotheliomatous meningioma, fibrous meningioma, and anaplastic meningioma have been reported to date.[1–8] The MRI findings in our case showed a gross variability in signal intensities between the two lesions in all sequences and even on contrast administration (►**Fig. 1**). The possible explanation for the variable MRI appearance could be due to the cell density and heterogeneity of the tumor type. The histopathological examination in our case showed two different histological patterns of the same grade (WHO grade 1)—transitional and angiomatous meningiomas. The exact mechanism of multicentricity is unknown.[9] The commonly recognized theory postulated that the initial cell clone disperses across the meninges, resulting in the development of several, clonally related tumors.[4,9] The second hypothesis, which is backed by the observed histological and cytogenetic variations among the several tumors from the same patient, postulates that tumors develop independently.[4,9] A recent comprehensive study using an scRNA-seq analysis of the human dura and primary meningioma tumor samples revealed the role of the dura in central nervous system (CNS) immune surveillance; the study also found copy number variant heterogeneity and heterogeneity in gene expression across different types of meningiomas.[10,11] The treatment protocol for different histological types in the same patient is not clear. A WHO grade 1 meningioma requires only surgical resection, whereas a WHO grade 2 meningioma requires surgery ± radiotherapy. However, the extent of surgical resection remains the mainstay of the treatment. In our case, the patient underwent a Simpson grade 1 excision of the lesion and was advised for close follow-up.

Conclusion

The advent of computed tomography (CT) and MRI has led to a gradual increase in the diagnosis of MMs, but MMs with different histologies are very rare and the exact mechanism is unknown. Intratumoral heterogeneity, being extensively researched by gene sequencing and molecular analysis, is yet to answer the question. The extent of surgical resection remains the mainstay of the treatment and these patients should be followed up closely to watch for recurrence or malignant transformation. The role of radiotherapy in MMs is yet to be established.

Conflict of Interest

None declared.

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