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Case Report

Microcystic Meningioma: Atypical Meningioma Revisited. Rare Case Report with Review of Literature

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Abstract

Meningiomas, classical extra-axial dural-based tumors of the meninges, have well-documented classical imaging features and signs on CT and MRI. Most meningiomas are classical lesions with prompt diagnosis on imaging and generally exhibit benign outcomes with slow growth patterns. The latest WHO classification of tumors in 2021 has classified 15 subtypes of meningiomas, and while classical meningiomas are WHO Grade 1 tumors, grading between varying subtypes varies between WHO Grades 1 and 3. The microcystic meningioma is a rare and atypical subtype of meningioma that has been sparsely documented in literature. In this case report, we revisit this rare subtype of meningioma with a brief review of literature.

More Information

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Introduction

Classical meningiomas are dural-based, extra-axial lesions representing the most common tumor of the meninges, having hallmark features and signs that have been well documented and rarely pose a diagnostic dilemma and generally have a benign, slow-growing pattern. The 5th WHO Classification of Brain Tumors, introduced in 2021, has classified meningiomas into 15 subtypes [1]. Knowledge of these separate subtypes is imperative as these lesions do not follow the imaging and clinical course associated with the classical meningiomas. Depending upon the subtype, the meningiomas are classified between WHO Grades 1 and 3. The Microcystic meningioma is a rare subtype of meningioma accounting for 1.6% of cases of intracranial meningiomas [2] that has been sparsely documented in the literature. This variant of meningioma has atypical imaging appearances and poses a diagnostic challenge; hence, knowledge of its imaging characteristics is essential to making a prompt diagnosis and ruling out alternative diagnoses. This subtype was first classified and described in the WHO Classification of brain tumors in 1993 [3] and is mostly classified under the WHO Grade 1 [4]. The exact cause of microcystic meningiomas is not well known, but tests show changes like microcysts, networks, and empty spaces, along with two specific patterns seen in imaging. In the current literature review available, no incidence of any postoperative recurrence has been documented.

We hereby present a case of a recurrent microcystic meningioma in a 49-year-old female patient who was operated on previously for an intracranial space-occupying lesion that was histologically confirmed as a microcystic meningioma (WHO Grade 1) and had a tumor recurrence 5 years after the initial surgery.

Case report

A 49-year-old female patient presented to our hospital with complaints of progressive headache, vertigo, and visual disturbance in the right eye in the form of diplopia for the past 3 months. The patient exhibited progressive, insidious-onset weakness in the right upper and lower limbs for the past 2 months. The symptoms aggravated on exertion and viewing screens, reading, and any other visual activity. The ophthalmological assessment revealed a reduction in visual acuity in the right eye.

Neurological evaluation of the patient revealed weakness of right upper limb and lower limb muscles (mean 3/5 of right upper limb and 4/5 of right lower limb—MRC scale). The patient was oriented to time, place, and person. Blood investigations were within normal limits. Additional complaints of hypertension and hypothyroidism were present for which the patient had been on long-term medication. The patient also had a significant history of being a follow-up case of microcystic meningioma of the right

fronto-temporo-parietal lobes and was diagnosed initially in 2019. Craniectomy and gross total tumoral excision were done outside our institution in 2019, and the diagnosis was confirmed on histopathology. The postoperative period was uneventful with no significant complaints. Our institution performed CE-MRI as part of the clinical and diagnostic workup.

On MRI, an extra-axial lesion was observed along the right frontoparietal temporal lobes. The lesion was dural-based, demonstrating an enhancing dural tail (Figure 1). It was homogenously hyperintense on T2 (Figures 2,3), hypointense on T1 (Figure 4), and showing heterogeneous reticular mesh-like poor post-contrast enhancement (Figure 5). No obvious restriction on DWI or low values on ADC was observed. The lesion was causing mass effect on the adjacent brain parenchyma with a midline shift of 11 mm towards the left side; effacement of the ventricular system; and moderate adjacent peritumoral edema was present. Mass effect on the brainstem was also observed with a component of inferior transtentorial herniation and crowding at the foramen magnum. Apart from the previous craniectomy defect, we observed no underlying significant calvarial changes.

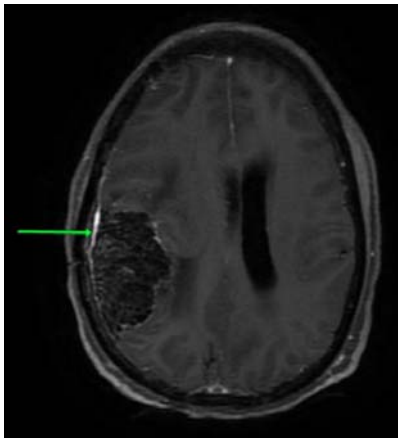

Figure 1: Axial Post contrast image shows an extra axial lesion with an enhancing dural tail (Green arrow).

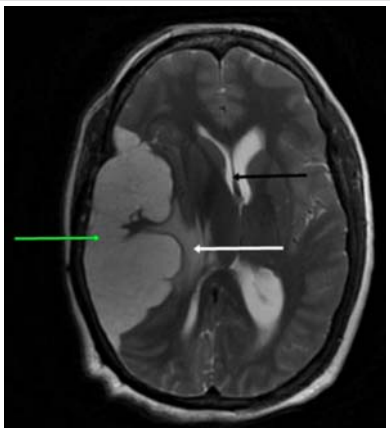

Figure 2: Axial T2 weighted image shows hyperintense lesion (Green arrow), Peritumoral edema (White arrow) and midline shift (Black arrow).

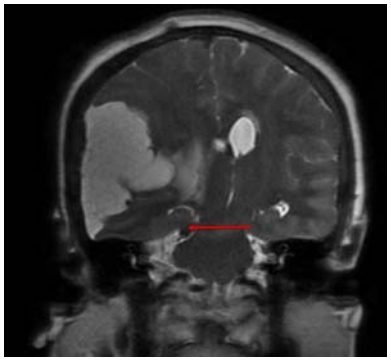

Figure 3: Coronal T2 weighted image demonstrated descending trans tentorial herniation secondary to mass effect from the lesion (Red arrow).

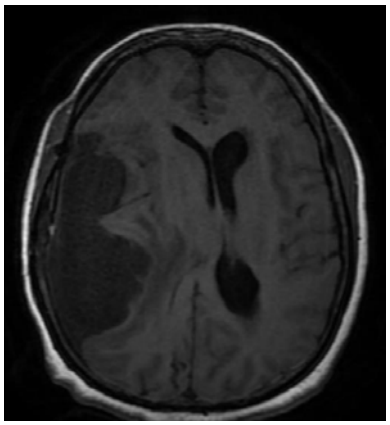

Figure 4: Axial T1 weighted image shows hypointense signal characteristics of the lesion.

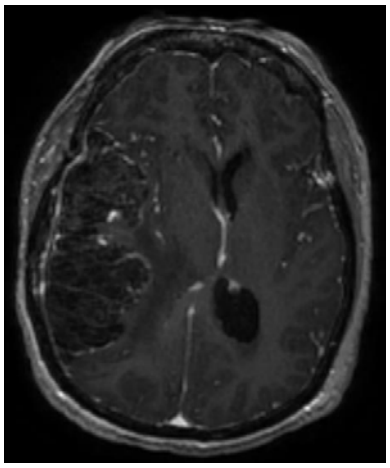

Figure 5: Axial T1 post contrast image shows fine mesh like reticular enhancement of the lesion.

Prompt surgical craniectomy was planned because of the significant midline shift and transtentorial herniation and to relieve visual symptoms secondary to compression of the optic chiasma. Gross tumoral excision was done, and the specimen was again sent for biopsy.

Histopathology showed cellular tumor tissue arranged in sheets and whorls with collagenous septa and was diagnosed as meningiothelial meningioma with microcystic subtype (Figure 6).

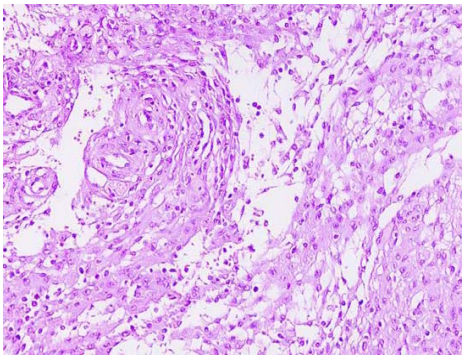

Figure 6: Tumor cells are round to oval with round nuclei, delicate chromatin, moderate amount of eosinophilic to clear cytoplasm and inconspicuous nucleoli. H&E with a high Ki-67 proliferation index. (20x).

The postoperative period was uneventful, and the patient was discharged on the 10th postoperative day and placed on follow-up.

Discussion

The various subtypes of meningiomas have uniquely different signal characteristics on MRI along with clinical behaviour and growth patterns, and hence, MR signal characteristics of meningiomas gives an important clue to the histological diagnosis. On review of literature, we observed that now a consensus exists regarding the signal characteristics of microcystic meningioma, i.e., the lesion appears hypointense on T1 and hyperintense on T2 with two separate patterns of post-contrast enhancement [7-9]. The lesion in our case was showing homogenous hyperintensity on T2, which on retrospective evaluation proved to be significant as the lesion was diagnosed as meningothelial meningioma, a finding considered statistically significant in independent studies by Elster, et al. [10] and Kaplan, et al. [11]. CJ Chen, et al. [8] in their study documented the importance of T1 hypointense signals seen within the lesion with a high confidence interval of 95% in differentiating classical meningiomas vs. microcystic subtype.

Seung Hyun Kim, et al. [7] demonstrated two separate enhancement patterns in microcystic meningioma: Pattern 1-homogeneous post-contrast enhancement like that observed in typical meningiomas and Pattern 2-homogeneously low or no post-contrast enhancement with mesh-like reticular areas of enhancement, as was observed in our case, although having a lesser incidence, was found to be highly specific (94%) by CJ Chen, et al. [8]. Similar observations have been reported by Willem Calderon, et al. [12] in their case report documenting a single case of microcystic meningioma.

Peritumoral edema is another imaging feature observed to be highly specific for this subtype [12] and was observed in our case. Kulanthaivelu, et al. [9] and Paek SH, et al. [13] also reported in their review of 28 and 16 cases respectively that vascular growth factors were the cause of the peritumoral edema and that higher expression of such factors was also responsible for the microcystic appearance of the lesion. CJ Chen, et al. [8] in their study reported a grading system of peritumoral edema based on the depth of edematous parenchyma adjacent to the lesion. According to this grading system, the parenchymal edema in our case was mild with a maximal parenchymal depth of 1.8 cm.

On review of the literature, we observed that no study exists documenting the recurrence of microcystic meningiomas. Classical meningiomas typically exhibit a recurrence rate between 0% - 2% for WHO Grade 1 lesions and 7% - 11% for WHO Grade 2 & 3 lesions [14]. The recurrence rates for WHO Grade I meningiomas after resection ranged from 7% to 23% over a 5-year period. Given that microcystic meningiomas are a subtype of WHO Grade I, it is plausible that their recurrence patterns align with those of other Grade I meningiomas [15].

Based on our observation of recurrence in a case of microcystic meningioma—and considering that WHO Grade 1 meningiomas can exhibit recurrence rates of up to 47% on long-term follow-up [16]— we propose that the microcystic subtype may exhibit a similar or potentially higher recurrence rate. This is further supported by focal areas of elevated Ki-67 expression identified on immunohistochemistry in our case. Barton, et al. [17] have demonstrated that a higher Ki-67 proliferative index is associated with an increased risk of recurrence in WHO Grade 1 meningiomas. Similarly, Kumar, et al. [18] reported a generally higher incidence of Ki-67 expression in microcystic meningiomas. Taken together, these findings suggest that the recurrence potential of microcystic meningiomas may be comparable to—or greater than—that of classical WHO Grade 1 tumors. Nevertheless, larger studies are needed to accurately document recurrence patterns in this uncommon histological subtype.

Conclusion

Meningioma is a commonly encountered extra-axial lesion seen in day-to-day practice and rarely causes any diagnostic challenge. Thorough knowledge of its various subtypes is necessary as they have distinct imaging characteristics and histological patterns, which translate to distinct tumor behavior, clinical progression, and outcomes. For the diagnosis of a microcystic variant of meningioma, an extra-axial lesion with imaging features such as T1 hypo-intensity, T2 hyperintensity, and characteristic enhancement patterns with peritumoral edema are the typical features.

Patient consent

The patient's written consent was obtained for the academic use and publication of her diagnostic images.

Ethical consideration

Waived off as the ethical committee did not consider this retrospective study to have any impact on the patient outcome.

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