



Case Report

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Anaplastic Meningioma Who Grade III: Case Report and Review of Immunohisto-pathological Characteristics

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Abstract

Introduction: Anaplastic meningiomas constitute about 2% of all intracranial meningiomas and are known to be aggressive with a mean overall survival and mean relapse-free survival of 3.3 years and 2.7 years respectively. We treated a patient with a World Health Organization (WHO) grade III Anaplastic Meningioma, a rare histological variant of convexity meningiomas in which intraoperatively we found the tumor mass was unique of mixed characteristics particularly peritumoral subdural effusion

Research Question: We show in this case report that convexity lesions presenting with subdural lesions should be highly suspected for WHO III anaplastic meningiomas backed with the immunohistopathological findings.

Material and Methods: Routine computed tomography for patients with neurologic signs and symptoms which reveal a convexity peri-tumoral subdural collection should be suspected for WHO III anaplastic meningiomas. Surgery and adjuvant radiotherapy offered a good clinical outcome.

Results: Postoperative the patient described a gradual improvement of the of expressive aphasia, deficits in speech, detectable personality changes or motor/sensory deficits. MRI at the three-month, six-month, one- year and two-year postoperative follow-up, revealed no evidence of residual tumor nor any observed recurrence nor recurrence of pre-operative symptoms.

Discussion and Conclusion In this paper, we present the rare radiological presentation and immunohistopathological characteristics of this WHO III Grade III anaplastic meningioma. WHO III anaplastic meningiomas should be considered in the differential diagnosis of intracranial tumors which present with peritumoral subdural collection on routine contrast tomography imaging. Our findings may contribute to the literature of the histopathological nature and behavior of this tumor subtype.

Keywords: Meningioma, anaplastic, WHO classification, peritumoral subdural effusion

Background Introduction

Anaplastic meningiomas constitute about 2% of all intracranial meningiomas and are known to be aggressive[1] with a mean overall survival and mean relapse-free survival at 3.3 years and 2.7 years respectively[2]. We treated a patient with a World Health Organization (WHO) grade III Anaplastic Meningioma, a rare histological variant of convexity meningiomas in which intraoperatively we found the tumor mass was uniquely of mixed characteristics particularly peritumoral subdural effusion. As the cyst walls did not adhere strongly to the underlying surrounding tissue, total extirpation of the tumor was successfully performed. We report

this case together with a pathological discussion. Ethical clearance was acquired from the Tongji Hospital Institutional Board Review to conduct the study.

Case Report History and Physical Examination

We describe a previously healthy 60-year-old male who was referred to our hospital in April 2016 with a complaint of severe worsening headaches. Prior to hospitalization, the patient had been admitted to local hospital complaining of dizziness and unstable gait for six days but with no headache. The local hospital cranial non contrast computed tomography (CT) scan showed left fron-

to-parietal subdural collection which was of mixed density with a high attenuation effusion initially misdiagnosed for an acute-on-chronic subdural hemorrhage and patient was hence planned for burr-hole craniotomy to drain the subdural collection. However, prior to surgery, the headache increased in intensity and patient was referred to our hospital for further investigation and definitive care.

At the time of presentation to our hospital the patient was complaining of severe headache. Review of systems was not suggestive of any recent changes within the past couple of months, including headaches, fatigue or changes in behavior. On physical examination he was conscious and oriented with bilateral and symmetric papillary response and intact extraocular muscle movements. Cranial nerves were otherwise intact. Neurological checkup of the

motor, sensory and cerebellar function was non revealing with no pathological findings. Preoperative laboratory workup revealed a normal basic metabolic panel, complete blood count, oncology panel and infectious panel.

Imaging

Head CT scan showed an extra-axial crescent shaped fluid collection over the left cerebral convexity indicating a chronic subdural hematoma and a high attenuation mass left parietal mass. Further magnetic resonance imaging (MRI) demonstrated a high signal 5.7x4.1 x2.9cm mass on T1 without a clear dural tail and a crescent shaped subdural vasogenic edema which caused a mass effect and a 10mm rightward sub-falcine shift as well as a compression of the frontal horn of the left lateral ventricle.

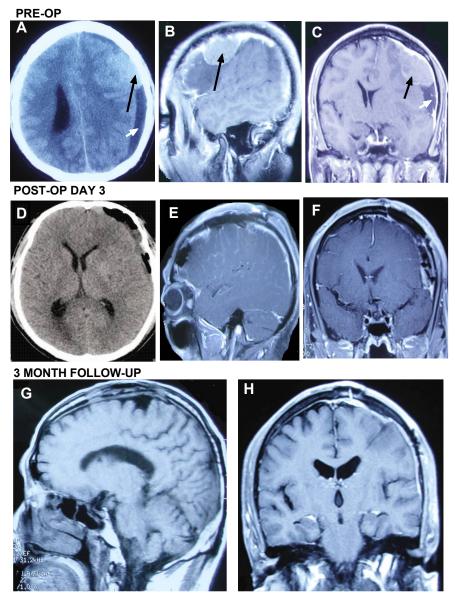


Figure 1: Preoperative, postoperative and follow-up images. A. Axial CT scan, B, C. MRI T1-Gd DWI scans demonstrating a convexity high attenuation mass (black arrow) with a midline shift to the right and peritumoral crescent shaped subdural fluid collection (white arrow). D, postoperative Axial CT. E, F postoperative MRI T1-Gd scans showing total resection and complete resolution of subdural effusion. G, H MRI T1-Gd scans at 3 months follow up revealed no evidence of residual tumor and no observed recurrence.

Treatment and Follow up

Symptomatic treatment was administered and the patient was kept under observation as the imaging examinations were completed. The primary goal of treatment in this case was surgical resection.

Histological examination of the resected tumor tissue revealed an anaplastic meningioma (WHO grade III) [FIGURE 2] and Immu-

nohistochemistry Assay revealed: Epithelial membrane antigen EMA (+) Vimentin VIM (+) protein S-100 (-) Desmin (+) SMA (-) Hematopoietic cell antigen

CD34 (-) Signal transducer and activator of transcription 6 STAT6 (-) Ki-67 (LI $\sim 50\%$)

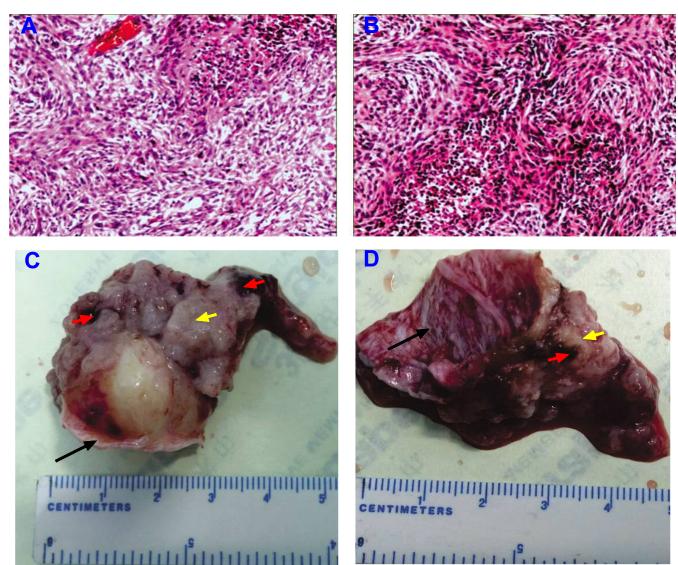


Figure 2: Histology and tumor tissue section. A, B H&E staining showing abundant mitoses, nuclear atypia and cellular pleaomorphism indicating anaplastic meningioma (H&E x 200). C, D sections of tumor tissue showing the dural attachment (black arrow) and necrotic grey soft mass(yellow arrow) with fresh hematoma(red arrow)

Postoperatively, the patient had no evidence of expressive aphasia, deficits in speech, detectable personality changes or motor/sensory deficits. Postoperative imaging showed no evidence of residual tumor, hemorrhage or infarction. He was discharged on postoperative day 8.

The patient received adjuvant chemotherapy and radiotherapy two weeks after discharge. At 3-month follow-up, routine check-up revealed a normal neurological examination and without any deficits or prior dizziness and unstable gait. He was not taking any med-

ications at this time. Furthermore, MRI at the three-month, sixmonth, one-year and two-year postoperative follow-up revealed no evidence of residual tumor, complete resolution of edema nor observed recurrence, nor recurrence of the initial presenting signs and symptoms.

Discussion

Anaplastic meningiomas with designation WHO grade III are generally reserved for lesions with histological evidence of malignancy including nuclear atypia and brisk mitotic activity [1]. These in-

frequent meningiomas have tissue characteristics and histological findings very distinct from other classifications of meningiomas although exhaustive histopathological definitions of this subtype remain to be well defined.

In this paper, we discuss the pathological characteristics of a rare form of WHO Grade III anaplastic meningioma. Meningiomas, by nature, are known to originate from meningothelial cells [3]. This particular case presented an unusual diagnostic picture different from other convexity subtypes of meningiomas given its imaging characteristics and intraoperative findings. Liu and colleagues suggested a high correlation between preoperative imaging characteristics of meningiomas and their actual diagnosis [4]. However, the imaging features of this case were not predictably comparable to common findings with other convexity meningiomas, particularly the peritumoral subdural hemorrhage observed fresh bleeding intraoperatively. By contrast, the physical signs and symptoms were not as aggressive as compared to the known malignant tendencies of other documented WHO grade III tumors [1]. Kim and colleagues in a case report reported that perioperative images of disproportion between aggressive peritumoral edema and tumor size were suggestive of secretory meningioma (WHO grade 1) [5] as much opposed to the pathology diagnosis of a WHO grade III anaplastic meningioma. Other intracranial tumors that present with this phenomenon include inclusions or cytoplasmic vacuolization seen in metastatic from breast ductal carcinoma, hepatocellular carcinoma or gastric carcinoma [5, 6]. The exact pathophysiologic mechanism of the development of the subdural peritumoral edema and the bleeding

within the tumor remains unclear, however, several hypotheses have been put forward which include and not limited tumor volume, progesterone hormones, interleukin-6 expression, possible involvement of mast cell-derived histamine or serotonin [7].

Immunohistochemically, the tumor cells were positive for epithelial membrane antigen (EMA) and vimentin (VIM), desmin, and negative for protein S-100, SMA, CD34, and STAT6. Ki-67(MIB-1; 1: 100 dilution) proliferation index was 50% .

However, the immunohistologic features of meningioma may not exactly reflect the histological subtype. Studies on EMA expression have reported conflicting results and has been described as a common but inconsistent finding by some researchers [8, 9], although the positive EMA in this case study excludes it from hemangiooericytoma which may mimick the aggressive behavior similar to anaplastic meningioma when inside the nervous system[10]. In this current case study, the immunohistology assay was negative for S-100 in contrast to a study comparing immunohistochemical profile of meningiomas by Andreas et al [9] in which four out of the five cases of anaplastic meningioma tested positive on immunostaining.

The proliferation index at 50% was consistent with previous studies[11, 12] in which meningiomas and other brain tumors showed good correlation with anaplasia or malignancy. Studies of Ag-NOR counts of primary tumors and recurrences according to their

clinical course and histopathological subtypes by Maier and Dietmar[10] reported a highly significant correlation with the labeling indices of Ki-67 in their three-step scale of classic, atypical and anaplastic meningiomas.

Conclusion

WHO III anaplastic meningiomas should be considered in the differential diagnosis of intracranial tumors which present with peritumoral subdural edema on preoperative CT imaging. Inasmuch as there is no consensus on the immunohistochemical markers is specific for diagnosis of particular this subtype, our findings may contribute to the literature of the histological nature and behavior of this tumor subtype.

Abbreviations

WHO - World Health Organization CT scan- computed tomography scan MRI - magnetic resonance imaging Epithelial membrane antigen EMA VIM - Vimentin SMA - smooth muscle actin CD3 Cluster of differentiation 3

STAT - Signal transducer and activator of transcription
Ag-NOR - Silver stained Nucleolar organiser regions Declarations

Ethics Approval and Consent to Participate

Ethical clearance was acquired from the Tongji Hospital Institutional Board Review to conduct the study.

Consent for Publication

Consent was acquired from the patient regarding publication of the case report. The document will be provided upon request from the editorial team.

Availability of Data and Materials

Data sharing is not applicable to this article as no datasets were generated or analysed during the current study.

Competing Interests

The authors declare that they have no competing interests.

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Authors' Contributions

The individual contributions of authors to the manuscript should be specified in this section. Guidance and criteria for authorship can be found in our editorial policies.

We suggest the following kind of format (please use initials to refer to each author's contribution): JL assisted in the surgery, participated in the pre and post-operative management and drafted the manuscript. KS carried out the surgery, participated in the pre and post-operative management and reviewed the manuscript. WJ KS carried out the surgery, participated in the pre and post-operative

management and reviewed the manuscript. TL conceived of the study, and participated in its design and coordination and helped to supervise and review the manuscript. All authors read and approved the final manuscript.

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