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Case Report

Diagnostic approach to a rare case of anaplastic meningioma with osteosarcomatous differentiation and associated bone alterations

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ABSTRACT

Anaplastic meningioma with osteosarcomatous differentiation is a very rare finding. We herewith present squash cytology, histopathology and immunohistochemistry findings of this rare case with systematic approach to diagnosis. A 38-year-old female presented with complaints of headache, vomiting, seizure with loss of consciousness and left side weakness. Radiologically, there was a heterogeneous hyperintense likely extra axial densely calcified solid lesion measuring approximately 4.2×4.1×3.5 cm along right high frontal convexity compressing the adjacent brain parenchyma. Histology sections revealed fibro collagenous tissue, devitalized bone, multiple vascular spaces lined by fibrous septa with giant cells, along with spindle cell and round proliferation at one end with vague whorl formation. High mitosis, (>20/10 hpf) along with malignant lacy osteoid closely abutting the highly pleomorphic cells were indicative of a malignant spindle cell neoplasm with osteosarcomatous differentiation. Systematic approach, immunohistochemistry with involvement of all the specialities involved led to the correct diagnosis and management of patient.

Keywords: Anaplastic meningioma, Osteosarcoma, Radiology, Diagnosis, Differentials

INTRODUCTION

The tumors which share the morphological and immunophenotypic profile of meningothelial cells are known as meningiomas. They are usually benign tumors which are subjected to surgical removal. Intracranial location with Dural base lesion is radiological signature of these tumors. Secondary changes in meningiomas are well known with varied morphological variants for example chondroid, myxoid, lipomatous, angiomatous, and secretory. The World Health Organization (WHO) CNS5 recommends classification of meningiomas into grade 1, 2 and 3 on basis of defined criteria for both atypical and anaplastic.[1] This is a major alteration as previously few variants like papillary and rhabdoid were considered grade 3 on basis of morphology alone.[2] Anaplastic meningioma is a WHO grade 3 meningioma which is defined meningothelial tumor with 20 or more mitoses per 10 high-power microscopic fields (0.16 mm^2) and/or demonstrating a loss of distinctive features resulting in melanoma/carcinoma/sarcoma-like morphology.[3] We herewith report a case of anaplastic meningioma with osteosarcomatous differentiation. The challenges faced during diagnosis and step by step approach to the case.

CASE REPORT

A 38-year-old female presented with complaints of headache, vomiting, seizure with loss of consciousness and left side weakness.

On magnetic resonance imaging (MRI) brain contrast study T1 hypointense and T2 and FLAIR heterogeneously hyperintense likely extra axial lesion measuring approximately 4.2×4.1×3.5 cm along right high frontal convexity with compression adjacent brain parenchyma along with cortical buckling. Subtle area of T1 hyperintensity is seen within (Figure 1A). Mild to moderate perilesional edema is seen within. Patchy area of diffusion restriction is seen on DWI (Figure 1B). Multifocal area of blooming seen. On post contrast, the lesion heterogenous enhancement with associated enhancement along adjacent duramater.

On contrast enhanced computed tomography (CECT) brain large predominantly ill-defined densely calcified solid lesion with cystic areas noted in right high frontal lobe with extensive perilesional edema and mass effect over right cerebrum and right lateral ventricle (Figure 1C).

Radiological differential was meningioma/gliosarcoma.

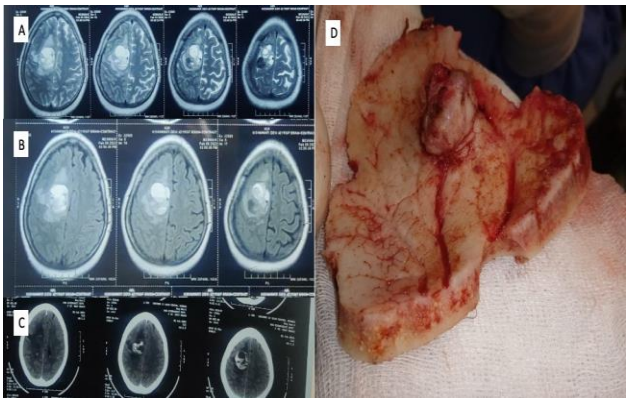

Figure 1: (A) Radiological image collage shows T1W1 image 4.2×4.1×3.5 along right high frontal convexity with compression adjacent brain parenchyma along with cortical buckling; (B) patchy area of diffusion restriction is seen on DWI; (C) on contrast CT the calcified solid lesion with cystic areas noted in right high frontal lobe with extensive perilesional edema and mass effect over right cerebrum and right lateral ventricle; and (D) the gross picture showed lesion attached to bone with smooth outer surface.

Pathological findings

Intraoperative squash cytology

Intraoperatively it displayed sheets of spindle cells with some areas of necrosis with questionable fibrillary material. It was reported as a high-grade lesion and further was deferred for definitive histology sections.

Definitive histology

On histological examination section showed multiple vascular spaces filled with blood. There intervening stroma show fibro collagenous tissue, devitalized bone, giant cells and fibrous proliferation along with mononuclear stromal at places (Figure 2). No glial tissue was identified. Occasional hemosiderin deposition also evident. So initially it was thought of an intraosseous bony lesion like aneurysmal bone cyst. However, at one end few cells having atypical morphology are evident closely opposed to osteoid (Figure 3B). This led to the morphological opinion of a primary bony lesion. By that time the radiology CT and MRI images were not seen by the reporting pathologist (PA).

Diagnostic approach

Firstly, the neurosurgical team was contacted to know about the complete clinical details and morphological opinion was discussed. On receiving the image of intra operative gross specimen and radiology (Figure 1A-D). It was confirmed that the lesion is primarily intracranial with dural attachment and bone changes not a primary intraosseous one.

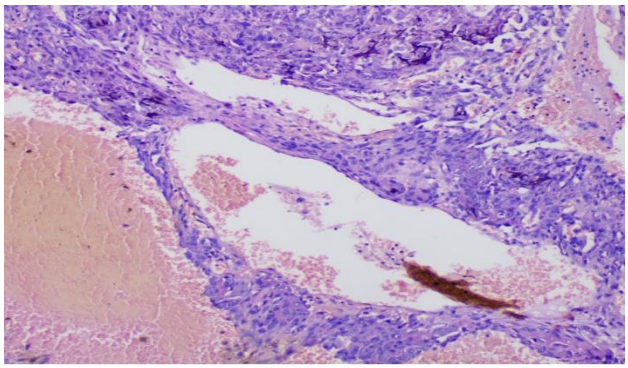

Figure 2: Microphotograph shows large vascular spaces lined by fibrous tissue along with scattered osteoclastic type giant cells (H&E x 100).

The morphological opinion was now directed to the spindle cells seen in one section (Figure 3a) where vague whorling was also appreciated. After careful observation the initial radiological diagnosis were again considered and immunohistochemistry for GFAP, vimentin, EMA and Ki67 were ordered. Mitosis was counted which were 22/10 hpf (Figure 3C).

Immunohistochemical results were GFAP was negative, vimentin was positive in tumor cells, EMA was positive in tumor cells and Ki67 was 10%. The immunohistochemistry was supportive of meningioma (Figure 4).

However, the tumor cells were seen abutting the thin lace like osteoid like material which was our second concern as it was something which could not be negated even after multiple reviews of the slides (Figure 3D). The published English literature was then sought for cases reported with osteosarcomatous differentiation in meningioma. After finding such report the diagnosis was dispatched. The

patient was provided with follow-up chemoradiation and is faring well after one year of follow-up.

So, to summarize the entire diagnostic line of reporting, the following points can be noted.

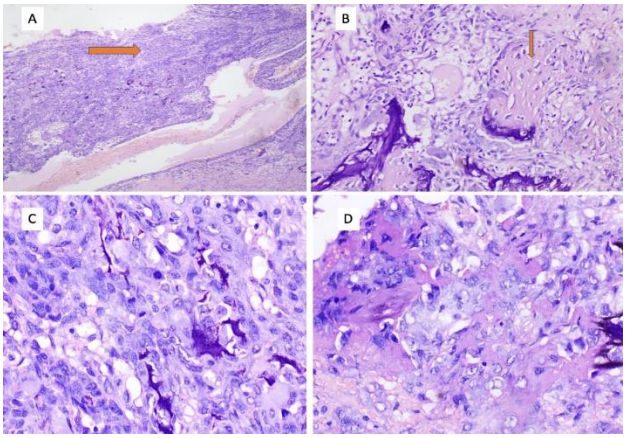

Figure 3: (A) Marked with arrow in scanner view areas with vague tumor cell whorling were evident as well (arrow; H&E x40); (B) areas of osteoid formation were also evident in processed tissue (H&E x200); (C) tumor cells display atypical nuclei with prominent nucleoli and brisk mitosis and; (D) atypical cells opposed to the lace like osteoid is well visualized in 3D (H&E x400).

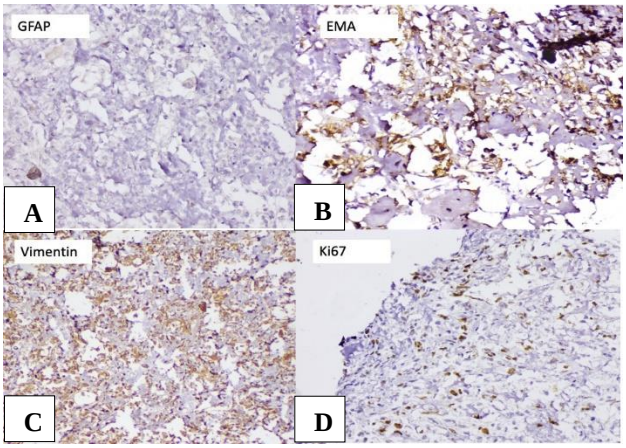

Figure 4 (A-D): Immunohistochemically tumor cells were negative for GFAP, and displayed EMA and vimentin expression with Ki67 of 20% (DAB x200).

On radiological review it was seen that the tumor was not originating from the bone per se, rather causing secondary bony changes this ruled out primary osteosarcoma.

10-15% Ki67 positivity suggested a high-grade lesion which was supported by both squash and histomorphological features and high mitotic count.

Immunohistochemistry showed GFAP negativity ruled out gliosarcoma.

EMA and vimentin positivity suggested dural origin tumor e.g. meningioma.

Additionally, >20 mitosis/10 hpf along with malignant lacy osteoid closely abutting the highly pleomorphic cells led to the diagnosis of secondary osteosarcomatous differentiation in anaplastic meningioma.

DISCUSSION

Anaplastic meningioma with osteosarcomatous differentiation is a very rare finding. On extensive search of literature, we found occasional case reports.[4,5]

Similar case was reported in 2022 from Japan where 82-year-old woman presented with motor aphasia. On MRI an extramedullary dural based tumor was seen in frontal region. They found multiple area areas with meningioma having mitotic count of 10-15/10 hpf reminiscent of grade 2 and areas where osteoid was seen closely abutting the tumor the mitotic count was >20/10 hpf which favoured grade 3 typing.[2] They had performed CDKN2A/2B gene study as well and observed the tumor had both hemi-deletion and had homozygous deletion of the CDKN2A/2B gene. According to them theirs was the index case and they stated that other allele was deleted in the CDKN2A/2B hemi deficient cells with benign morphology, and the homozygous deletion cells transformed into malignant transformation.

According to WHO CNS5 our case morphologically qualified to be grade 3 lesions with presence of high-grade sarcoma like features and high mitotic count. Radiological review was vital in the present case in establishing that the lesion was not primary osseous, and the large vascular areas which were reminiscent of aneurysmal bone cyst were secondary changes occurring due to the meningeal lesion. The microscopic evidence of vague whorl arrangement and high-grade tumor cells were present in only one section rest of the tumor areas had undergone so much calcification that the high-grade tumor was not evident in these areas. Hence careful observation of all sections and considering all viable areas is important for any differential. Lastly, immunohistochemistry was vital in summarizing the entire scenario.

CONCLUSION

The present case was diagnosed with collaborative effort of the radiological, neurosurgical and pathological inputs and it highlights how differentials can be worked out. The radiological evidence of the tumor being primary intracranial not intraosseous, backed by intraoperative specimens' resection features and finally supported by morphological features and immunohistochemistry led to the final diagnosis of this rare lesion.

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