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Case Report

Multifocal oligodendroglioma with callosal and brainstem involvement

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ABSTRACT

Background: Oligodendrogliomas are generally low-grade glial neoplasms commonly occurring in a cortical or subcortical location and frequently contain coarse calcifications. Tumors with 1p and 19q codeletions behave atypically and are more likely to have ill-defined margins and tend to have calcification. Very rarely, diffuse pattern and gliomatosis type of infiltrative nature of oligodendrogliomas have been described in sporadic case reports.

Case Description: In this article, we present a case of a 31-year-old male who had diffuse multifocal oligodendroglioma with rare features of extensive callosal and brainstem involvement on imaging.

Conclusion: Rare cases of oligodendrocytic gliomatosis cerebri or oligodendrogliomatosis with diffuse white matter spread of these tumors usually lead to a detrimental course of neurological status and a poor prognosis in these patients.

Keywords: Brain, Infiltration, Oligodendroglioma, Spectroscopy, Tumor

INTRODUCTION

Oligodendroglioma is glial neoplasms and they represent up to 5–20% of all gliomas.[10] The peak age in adults is between 40 and 60 years of age. Low-grade tumors are generally present in a younger age group than those with high-grade and anaplastic tumors. Although oligodendroglioma is sometimes considered relatively benign because of their initial indolent disease course, they are almost invariably fatal. Most oligodendrogliomas occur in the cerebral white matter, but they can be found anywhere in the central nervous system. In general, IDH mutation tumors are more likely to be found in the frontal lobe and extend across the midline more often compared to tumors that lack these deletions, which are more common in the temporal lobe and insula.[10,11] In this article, we present a case of a 31-year-old male who had multifocal oligodendroglioma with rare features of callosal and brainstem involvement on imaging.

CASE PRESENTATION

A 31-year-old male patient presented with a 3-month history of gradually worsening headache and progressive left-sided weakness. He denied any history of seizures or any other significant medical or surgical history. On examination, the patient had papilledema, left-sided upper

motor neuron type facial weakness, and left hemiparesis (MRC Grade 4) with pronator drift. Plantars were upgoing on the left side, suggesting complete lateralization. Rest of the examination was unremarkable.

Magnetic resonance imaging (MRI) revealed diffuse signal abnormality, multifocal lesions in the right frontal, temporal lobes with an involvement of the right thalamus, and extension into the right half of the brainstem up to the pons. Some of the lesions were cortical and subcortical in location and cystic changes were also seen in the tumor on T2-weighted imaging [Figure 1]. There was infiltration of the corpus callosum and extension to the contralateral left side and patchy lace-like enhancement on postcontrast T1-weighted images [Figure 2]. No diffusion restriction or susceptibility was noted. Along with MR spectroscopy (MRS) findings [Figure 3], a radiological diagnosis of low-grade multifocal glioma was made.

Biopsy was done and microscopy of the lesion revealed a tumor invading glial tissue composed of sheets of round cells with moderate, pale eosinophilic to clear cytoplasm, round to slightly oval vesicular nuclei, and predominantly inconspicuous nucleoli. Characteristic chicken-wire vasculature was present. Occasional scattered mitoses were seen without any endothelial vascular proliferation or necrosis. Tumor cells showed positivity for immunostain glial fibrillary acidic protein, occasional weak positivity for p53, and low Mib-1 (Ki-67) index (1–2%). Immunostain alpha-thalassemia/mental retardation syndrome, X-linked was intact. Based on these combined features, diagnosis of oligodendroglioma, not otherwise specified, WHO Grade II was favored [Figure 4].

No further surgical treatment was offered due to extensive involvement with a grave patient prognosis. A postbiopsy computed tomography (CT) scan was carried out because the patient had a sudden drop in his Glasgow Coma Scale. The CT scan showed diffuse brain edema and confirmed coarse calcifications within parts of the lesion [Figure 5]. Efforts were made to reduce the intracranial pressure with osmotic therapy, but the patient remained in a vegetative state.

DISCUSSION

Oligodendrogliomas are typically of low density on nonenhanced CT, low signal intensity on T1-weighted, and high signal intensity on T2-weighted MRI. The characteristic imaging features of oligodendrogliomas are the presence of coarse calcification (seen in up to 90% of patients), a cortical-subcortical location, heterogeneously increased signal intensity on T2-weighted sequences, and an indistinct border.[11] Minimal to moderate patchy and multifocal enhancement with a dot-like or lacy pattern is reported in up to 50% of tumors in patients with oligodendroglioma.

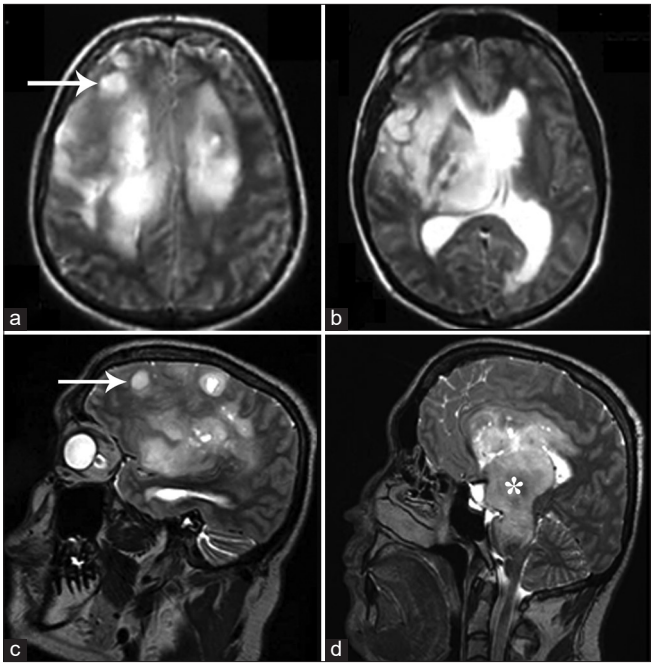

Figure 1: T2-weighted axial (a and b) and sagittal (c and d) images of magnetic resonance imaging brain showing multifocal T2 hyperintense lesions demonstrating mass effect, involving the thalamus, and extending into the brainstem (white asterisk in D). Some of the lesions were cortical and subcortical in location (white arrows in a and c) and cystic changes were also present.

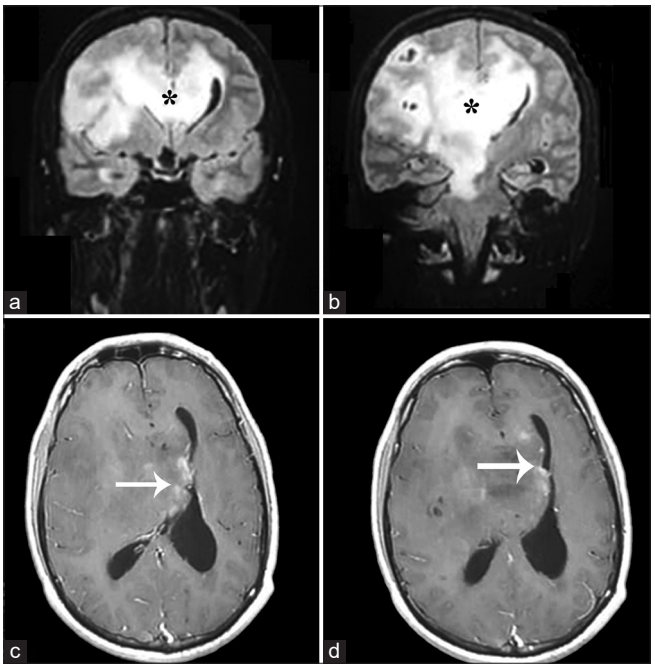

Figure 2: Coronal FLAIR (a and b) and postcontrast T1 (c and d) images of magnetic resonance imaging brain showing diffusely infiltrative lesion involving the corpus callosum (black asterisk in a and b) and extending to the contralateral side. Patchy lace-like enhancement was seen (white arrows in c and d); however, most of the lesions were largely nonenhancing.

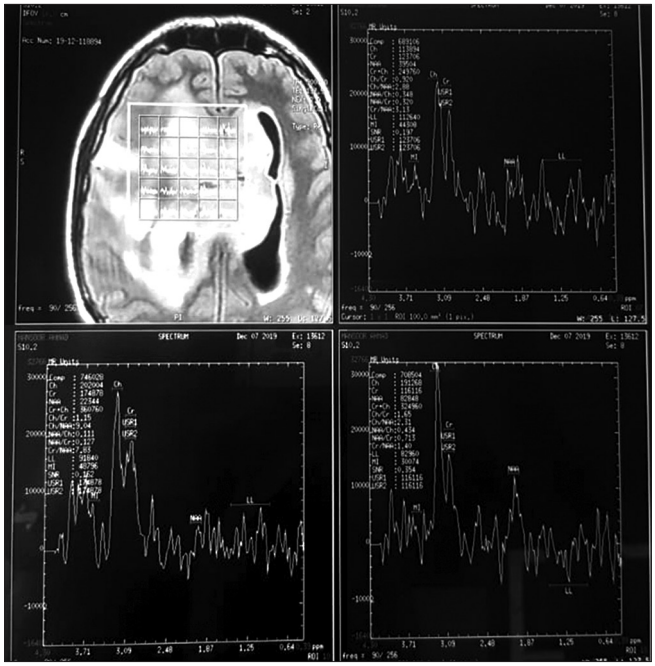

Figure 3: Magnetic resonance spectroscopy: Frontal lesion showed relative reduction in NAA compared to creatine and an increase in choline compared to creatine. NAA is a marker for normal neurons and axons, creatine is an internal standard and an increase in choline reflects an increase in cellular division. No raised lipid/lactate peak was noted.

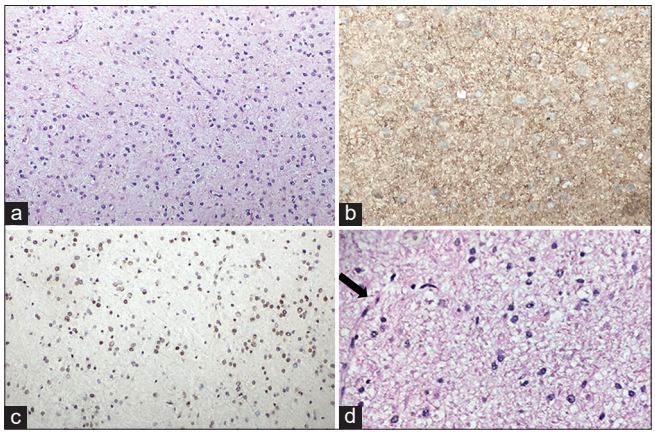

Figure 4: (a) Sheets of round cells with moderate eosinophilic cytoplasm and scattered thin blood vessels, H&E, 20×. (b) Cytoplasmic GFAP positivity, 20×. (c) Intact alpha-thalassemia/mental retardation syndrome, X-linked nuclear stain, 20×. (d) High-power view (H&E, 40×) showing characteristic chicken-wire vessel (black arrow).

Occasionally, frontal lobe gliomas may extend through the corpus callosum to produce a "butterfly glioma" pattern; however, this is more common in higher grade glioblastoma. There have been isolated callosal oligodendroglioma reports as well.[3,5] Tumors with 1p and 19q codeletions have noteworthy imaging features. They are also more likely to have ill-defined margins on T1-weighted MRIs and tend to have calcification. They are more likely to be found in the frontal lobe and extend across the midline more often compared to tumors that lack these deletions, which are more common in the temporal lobe and insula.[4] Due to its peripheral location, focal thinning, remodeling, or even erosion of the overlying skull are not uncommon in oligodendrogliomas. Infratentorial location is reported to be rare. The ventricular system, brainstem, spinal cord, retina, and leptomeninges are even rarer sites. Cystic degeneration, peritumoral edema, and hemorrhage may be found infrequently. Such features point toward anaplastic degeneration of the tumor.[7]

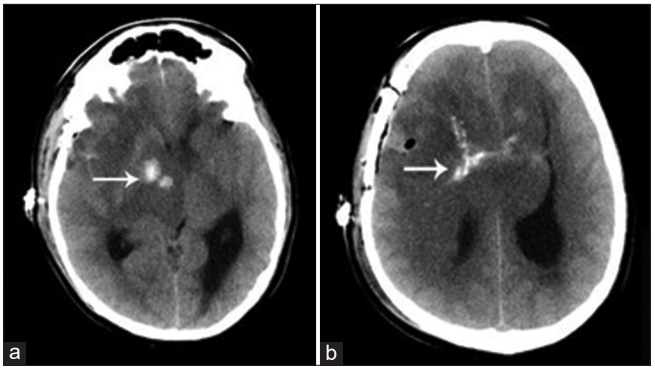

Figure 5: Plain computed tomography axial images (a and b) showing coarse calcifications within parts of the lesion (white arrows). Postcraniotomy and biopsy changes are also visible on the right side.

Although uncommon, the occurrence of multiple gliomas has been previously reported. They can be grouped in multicentric and multifocal depending on their mechanism of spread.[2] Multifocal gliomas disseminate from a primary site along nerve fiber bundles, cerebrospinal fluid channels, and blood. Multicentric gliomas consist of separate entities, in different lobes or hemispheres, and show no spread or metastasis. The exact incidence of multiple gliomas is unknown; a range between 0.5% and 20% has been quoted in the previous reports, with most reports describing two lesions in either the separate or the same lobe.[6,8,12]

Rare cases of oligodendrocytic gliomatosis cerebri or oligodendrogliomatosis have been reported in the literature, indicating white matter spread through the corpus callosum and brainstem, leading to a detrimental course of neurological status and a poor prognosis.[1,6,9] This pattern of involvement makes it a possibility in our case as well due to multiple lobar involvements; however, this diagnosis is usually made on autopsy. The limitation in our case report is the lack of molecular diagnosis because of financial constraints and guarded patient prognosis, which could have provided more insight into the IDH mutation and 1p and 19q genetic codeletions. However, this type of infiltrative pattern

is rarely seen in oligodendrogliomas and adds to the growing body of literature pertaining to neuro-oncology.

CONCLUSION

Multifocal low-grade oligodendrogliomas are uncommon and even though they are initially insidious in their clinical presentation, there is always a concern for potential malignant transformation. MRI and MRS, along with biopsy and histopathological correlation, are needed for a definitive diagnosis. The debate of surgery, neuro-oncological treatments (radiotherapy and chemotherapy) versus imaging surveillance for these types of cases, remains controversial. However, through this case report, we emphasize that radiologists and neurosurgeons should be mindful of such an infiltrative type of pattern of these tumors.

Declaration of patient consent

Institutional Review Board (IRB) permission obtained for the study.

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Conflicts of interest

There are no conflicts of interest.

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