Glioma Simultaneously Present with Adjacent Meningioma: Case Report and Literature Review

Abstract
A 51-year-old male patient presented to us with an episode of generalized tonic-clonic seizure. Magnetic resonance imaging revealed a dural-based contrast-enhancing lesion in the right temporal lobe and another heterogeneously contrast-enhancing intra-axial lesion in the right insula adjacent to it. Histopathology confirmed it as a meningioma adjacent to an anaplastic oligodendroglioma. This is only the second such case reported in literature. Literature on “adjacent site” gliomas and meningioma was also reviewed.

Keywords: Double tumors, glioma, meningioma

Introduction
Multiple primary brain tumors are usually seen in the presence of genetic disorders such as neurofibromatosis, tuberous sclerosis, or in patients who have received radiotherapy.[1] In the absence of such factors, multiple primary brain tumors, especially at adjacent sites, are rare. We present a case of a 51-year-old male patient who presented to us after a single episode of generalized tonic-clonic seizure and was finally diagnosed with “adjacent site” anaplastic oligodendroglioma and meningioma. This is only the second such case reported in literature.

Case Report
A 51-year-old male presented to us with a complaint of one episode of generalized tonic-clonic seizure 15 days ago. The seizure was associated with loss of consciousness for 10 min and jerky movements of all four limbs. There was no history of aura, postictal headache or weakness, tongue bite, or loss of continence associated with it. On examination, he had no neurological deficit. There were no neurocutaneous markers. Magnetic resonance imaging brain [Figure 1] revealed an ill-defined area of altered signal intensity in the right frontotemporal and insular region which was hypointense on T1, hyperintense on T2 with patchy areas of enhancement suggestive of a high-grade glioma. Another avidly enhancing lesion dural-based extra-axial lesion was overlying right temporal lobe (adjacent to the other lesion) suggestive of a meningioma.

Under neuronavigation, the patient underwent the right frontotemporal craniotomy. After reflecting the dural flap anteroinferiorly, a dural-based lesion was seen attached to the temporal dura and extending into sylvian fissure suggestive of meningioma [Figure 2]. The sylvian fissure was opened up due to lesion. The other tumor was intra-axial, soft, grey-white, suckable, moderately vascular, and extended into frontal operculum and temporal lobe around the right middle cerebral artery suggestive of high-grade glioma. Postoperative period was uneventful, and the patient was discharged on postoperative day 6. On histopathological examination [Figure 3], the dural-based lesion revealed a tumor composed of sheets and whorls of spindle cells. There was mild nuclear atypia. Few psammoma bodies were also present. These findings were suggestive of Grade 1 meningioma. The intra-axial lesion revealed a moderately cellular tumor composed of sheets of tumor cells with a fibrillary background. There was moderate nuclear atypia with increased mitotic activity. On immunohistochemistry, tumour cells were positive for glial fibrillary acidic protein, p53. Ki-67 was 12%–15%. The findings were suggestive of anaplastic oligodendroglioma (Grade 3).

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Discussion
Multiple intracranial tumors are extremely rare. They are more commonly seen in patients with genetic disorders such as neurofibromatosis or tuberous sclerosis. They may also be seen in patients who have received radiotherapy treatment. The incidence of primary brain tumors with different histology is 0.3% of all brain tumors. The most frequently occurring combination is that of meningioma and glioma, followed by meningioma and neurinoma and meningioma and pituitary adenoma. Even though meningioma along with glioma is the most frequently occurring combination, its occurrence at adjacent sites has been reported by only a few authors [2-10] [Table 1]. More so, occurrence of an oligodendroglioma adjacent to a meningo- glioma is even rarer, this case being only the second such case available in the English literature [2].

Various hypotheses have been proposed to explain the occurrence of this rare phenomenon. Since meningiomas and gliomas are the most commonly occurring primary brain tumors, most authors feel that this concurrence of meningioma and glioma at adjacent sites is a mere co-incidence. Certain genetic factors, exposure to chemicals, trauma, or some immunological mechanism may be responsible for this phenomenon. Some are of the opinion that a locally acting oncogenic paracrine factor from meningioma may induce malignant transformation in adjacent brain parenchyma and glial cells. Similarly, irritative effects from low-grade glioma may induce meningeal proliferation. Exact mechanism is, however, yet to be elucidated.

In such cases, it is usually not possible to make a preoperative diagnosis of “double tumors.” One must be aware of the possibility of such an entity and suspect it if perilesional edema does not correlate with the presence of

| Author (et al.) | Year | Age (years) | Sex (male/female) | Location | Pathology |
|----------------|------|-------------|-------------------|----------|-----------|
| Gass and Van Wagenen | 1950 | 56 | Female | Right frontal | ODG + meningioma |
| Nagashima et al. | 1963 | 42 | Male | Left frontal | Glioma (low grade) + fibroblastic meningioma |
| Strong et al. | 1976 | 56 | Female | Right frontal | GBM + meningioma |
| Strong et al. | 1976 | 53 | Male | Lt. frontal | GBM + syncytial meningioma |
| Marra et al. | 1977 | 63 | Male | Right parietal | GBM + meningothelial meningioma |
| Goyal et al. | 2003 | 72 | Male | Right temporal | GBM + fibrous meningioma |
| Nestler et al. | 2007 | 49 | Male | Left frontal | GBM + fibrous meningioma |
| Suzuki et al. | 2010 | 75 | Female | Left temporal | GBM + meningothelial meningioma |
| Chen et al. | 2010 | 63 | Female | Left frontal | GBM + fibroblastic meningioma |
| Shahmohammadi et al. | 2016 | 49 | Female | Right frontal | GBM + meningothelial meningioma |
| Present case | 2017 | 51 | Male | Right temporal | Anaplastic ODG + meningioma (Grade I) |

ODG – Oligodendroglioma; GBM – Glioblastoma multiforme
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a single benign lesion or intraoperatively the tumor does not correlate with preoperative radiology. In certain cases, perilesional edema in meningioma may mask a small low-grade astrocytoma. Biopsy helps to provide tissue diagnosis in such cases.

**Conclusion**

Multiple primary brain tumors are rare in the absence of predisposing factors such as genetic disorders or radiotherapy. A surgeon must be aware of the entity “adjacent site” double tumors when radiological and intraoperative findings do not corroborate. Biopsy helps to confirm the diagnosis in these cases.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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**Conflicts of interest**

There are no conflicts of interest.

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![Figure 3](image-url)