Case Report

Anaplastic oligodendroglioma presenting with apoplectic intratumoral hemorrhage✩

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ARTICLE INFO

Article history:
Received 28 October 2022
Accepted 2 November 2022

Keywords:
Intratumoral hemorrhage
Apoplectic
Anaplastic oligodendroglioma
Genomic analyses

ABSTRACT

A 31-year-old woman presented with a headache and nausea. At presentation, her blood pressure was 114/71 mm Hg with left hemiparesis. Computed tomography revealed a large hyperdense mass in the right temporal lobe accompanied by intraslesional calcifications and ventricular perforation. Spot signs were not identified, and cerebral angiography did not reveal any abnormal vasculature. The patient underwent emergency craniotomy assuming an intracerebral hemorrhage. Intraoperatively, grayish tumor tissue was found to intermingle with the clots. Microscopic findings of the tumor revealed neoplastic cells possessing perinuclear halo and cell atypia, and diffusely stained with glial fibrillary acidic protein, which were consistent with anaplastic oligodendrogliomas. However, genomic analyses of the tumor showed non-mutant isocitrate dehydrogenase 1 and telomerase reverse transcriptase, in addition to wild-type O6-methylguanine DNA-methyltransferase. These are equivalent to glioblastoma multiforme. Based on the results, we assumed that anaplastic oligodendrogliomas may develop apoplectic intratumoral hemorrhages that mimic intracerebral hemorrhage. Genomic exploration is recommended for such tumors, coupled with careful follow-up, owing to its potentially aggressive nature.

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✩Competing Interests: The authors have declared that no competing interests exist.
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https://doi.org/10.1016/j.radcr.2022.11.017
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Introduction

Apoplectic intratumoral hemorrhages are rare entities and they most-frequently develop in pituitary macroadenomas, such as pituitary apoplexy [1–3]. Except for pituitary adenomas, such hemorrhages have been documented to occur in glioblastoma multiforme, astrocytoma, oligodendrogloma, anaplastic oligodendrogloma, ependymoma, malignant lymphoma, pleomorphic xanthoastrocytoma, pilocytic astrocytoma, hemangioblastoma, plasmaeyctoma, and metastatic carcinoma [4–16]. In an autopsy series of 430 spontaneous intracerebral hemorrhages, 10.2% were caused by neoplasms [17].

Recently, adult-type diffuse gliomas have been broadly categorized into three types: isocitrate dehydrogenase (IDH)-mutant astrocytoma, IDH-mutant and 1p/19q codeleted oligodendrogloma; and IDH wild-type glioblastoma multiforme [18].

Here, we present a unique case of anaplastic oligodendrogloma presenting with an apoplectic intratumoral hemorrhage that was genetically equivalent to glioblastoma multiforme.

Case report

A 31-year-old previously non-hypertensive woman presented with an abrupt headache and nausea during desk work. The patient was urgently transported to the hospital in the middle of the night. At presentation, her blood pressure was 114/71 mmHg, motor paresis was detected in the left upper and lower extremities, and the manual muscle test score was 2/5. Computed tomography (CT) revealed a large, hyperdense mass in the right temporal lobe. It measured 45 × 34 mm and was accompanied by perilesional brain edema, intralesional calcifications, and ventricular perforation (Fig. 1). On contrast CT, no spot signs were identified in the hemorrhage (Fig. 2A). Cerebral angiography did not reveal any abnormal vasculature or arteriovenous shunts (Fig. 2B). The patient underwent an emergent craniotomy, assuming an intracerebral hemorrhage caused by cerebrovascular disease. However, intraoperatively, grayish tumor tissue was found to intermingle with the clots. The tumor which was elastic soft in consistency and less vascular, was subtotally resected. Microscopically, the tumor comprised of neoplastic cells possessing perinuclear halos and cellular atypia. They were diffusely stained for glial fibrillary acidic proteins with the MIB-1 index approximately 10% (Fig. 3). The microvascular proliferation was less prominent and lacked necrotic foci, and using fluorescence in situ hybridization, codeletion of 1p/19q was not identified. These findings were consistent with those of anaplastic oligodendrogloma, a world health organization (WHO) grade 3 tumor. In contrast, genomic analyses of the tumor revealed non-mutant IDH 1 and telomerase reverse transcriptase, in addition to wild-type O6-methylguanine DNA-methyltransferase. These were equivalent to WHO grade 4 glioblastoma multiforme (Fig. 4). The patient’s postoperative course was uneventful, with complete resolution of her neurological impairments. Following adjuvant chemoradiation therapy, the patient was under close observation by periodical magnetic resonance imaging.

![Fig. 1: Axial computed tomography scan showing a large hyperdense mass in the right temporal lobe (A, asterisk), 45 × 34 mm in dimension, with perilesional brain edema, intralesional calcifications (dashed arrows), and ventricular perforation (B, asterisk).](image-url)
Fig. 2 – Postcontrast axial computed tomography scan (A) and lateral view of the internal carotid angiography (B) showing no spot sign in the hemorrhagic lesion (A, asterisk) or abnormal arteriovenous shunts.

Fig. 3 – (A) Photomicrograph of the resected specimens showing tumor tissue comprising of neoplastic cells with perinuclear halo and cell atypia. (B) Photomicrograph of the MIB-1 index accounted to be approximately 10%. (A) Hematoxylin and eosin stain, ×400; (B) ×400.

Discussion

Apoplectic intratumoral hemorrhage is a rare occurrence among brain tumors. If encountered in an emergency setting, surgery for hematoma evacuation can be erroneously planned, resulting in incomplete tumor resection and loss of actual diagnosis. The present case was that of a non-hypertensive young woman. Furthermore, the identified hematoma was accompanied by intrallesional calcifications. Intratumoral hemorrhage should be assumed in the differential diagnosis of patients with intracerebral hemorrhage with atypical clinical appearance. Although high-grade malignancy and extensive tumor vessels have been proposed as predisposing factors for such hemorrhages, low-grade, less-vascular oligodendrogliomas have been reported to cause the hemorrhages [5,7–11,13,17]. As primary brain tumors presenting with apoplectic intracerebral hemorrhage, low-grade oligodendrogliomas are a frequent entity [7,8]. In contrast, there have been few documented cases of high-grade oligodendroglioma causing apoplectic hemorrhage [6]. In the present tumor, diagnosis based on morphological and immunohistochemical explorations was consistent with less vascular WHO grade 3 anaplastic oligodendroglioma. However, it was equivalent to WHO grade 4 glioblastoma multiforme, based on the recent classification of diffuse gliomas [18]. Apoplectic intratumoral hemorrhage in oligodendrogliomas may reflect its more aggressive nature than non-hemorrhagic ones. To elucidate the precise mechanism and contributing factors of such hemorrhages in oligodendrogliomas, molecular biological approaches coupled with case accumulation and long-term follow-up would be critical.

Anaplastic oligodendrogliomas may develop apoplectic intratumoral hemorrhage that mimics intracerebral hemor-
Fig. 4 – Genomic analyses of the resected specimens showing non-mutant isocitrate dehydrogenase 1 (A) and telomerase reverse transcriptase (B), in addition to wild-type O6-methylguanine DNA-methyltransferase (C), equivalent to glioblastoma multiforme. IDH, isocitrate dehydrogenase; MGMT, O6-methylguanine DNA-methyltransferase; TERT, telomerase reverse transcriptase.

rhage. For tumors of a potentially aggressive nature, genomic exploration coupled with careful follow-up is recommended.

Patient consent

The patients documented in the manuscript fully understood and agreed that the authors use the information materials of the patients in anonymized manner for possible publication in Radiology Case Reports.

Author contributions

All the authors contributed equally to this study.

Ethical standards

We declare that the present study has been approved by the institution’s guidelines for human research and was performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

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