Case Report

Cystic Glioblastoma Rupturing into the Ventricle

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Cystic tumors, such as craniopharyngiomas and Rathke's cleft cysts, as well as arachnoid cysts have been reported to rupture occasionally. Approximately 8-10% of glioblastomas (GBMs) are known to have a significant cystic component; however, to the best of our knowledge, no studies have reported cystic rupturing of GBMs. Here, we describe a unique case of cystic GBM rupturing and penetrating into the cerebral ventricle. A 77-year-old man with a right frontal lobe lesion suspected as GBM with a large cyst was referred to our hospital. At admission, disorientation and left facial weakness were detected. Consciousness disturbance worsened on the 8th day of hospitalization. Computed tomography (CT) revealed prominent shrinkage of the tumor and intratumoral cyst. Signs of meningeal irritation were observed, and chemical meningitis due to cystic tumor rupture and leakage of necrotic components into the ventricle was highly suspected. Surgical resection of the right frontal lobe tumor was performed on the 10th day of hospitalization. During the surgery, clear and colorless cerebrospinal fluid was obtained upon penetration of the tumoral cyst, suggesting traffic of tumor cysts and cerebral ventricle. Adjuvant chemoradiation therapy was initiated postoperatively. Local recurrence was noted at the corpus callosum 7 months postoperatively and was treated with a gamma knife. Further therapy was performed after this recurrence. However, his condition gradually deteriorated 15 months postoperatively, and he was subjected to terminal care. To the best of our knowledge, this is the first report on a cystic GBM rupture.

Keywords: glioblastoma, cyst, rupture, ventricle

Introduction

Glioblastomas (GBMs) represent glial tumors with a high proliferative activity, high occurrence rate, and tendency for destructive infiltration; therefore, they are clinically one of the most malignant tumors. They are known to rapidly progress with intratumoral hemorrhage and cyst formation. The presence of a cyst has been reported in approximately 10% of the central nervous system tumors.¹⁾ Cystic tumors, such as craniopharyngiomas and Rathke's cleft cysts, have been reported to rupture occasionally.^{2,3)} However, to the best of

Received: March 3, 2019; Accepted: July 10, 2019 Online December 18, 2019

Copyright© 2020 by The Japan Neurosurgical Society This work is licensed under a Creative Commons Attribution-NonCommercial-NoDerivatives International License. our knowledge, no studies have reported cystic tumor rupture of GBMs.

Here, we report a rare case of GBM rupturing and penetrating into the cerebral ventricle.

Case Presentation

A 77-year-old man who developed walking disturbance and urinary incontinence visited a local doctor. Head magnetic resonance imaging (MRI) revealed a right frontal lobe lesion suspected as a GBM, and the patient was referred to our hospital. Disorientation and left facial weakness were detected at the time of admission. MRI with contrast enhancement on admission revealed a neoplastic lesion with a large cystic structure at his right frontal lobe (Figs. 1 and 2A). Elective surgery was planned as GBM was suspected. On the 8th day of hospitalization, 2 days before the planned surgery date, consciousness disturbance worsened to E3V4M5 on the Glasgow Coma Scale. Computed tomography (CT) was performed because tumor enlargement, edema progression, or intratumoral hemorrhage was suspected (Fig. 2B), which revealed prominent shrinkage of the tumor and intratumoral cyst. Signs of meningeal irritation were also observed. Chemical meningitis due to cystic tumor rupture and leakage of necrotic component into the ventricle was highly suspected (Fig. 2C). Therefore, we initiated treatment with dexamethasone and glycerol. On the 10th day of hospitalization, surgical resection of the right frontal lobe tumor was performed. During the surgery, clear and colorless cerebrospinal fluid was obtained upon penetration of the tumoral cyst, suggesting traffic of tumor cysts and cerebral ventricle. Examination of this cerebrospinal fluid revealed 19 cells per microliter, three mononuclear cells per microliter, seven polynuclear cells per microliter, seven difficult to classify cells per microliter, 7 mg/dL glucose, and 4202 mg/dL protein. Excision along the tumor boundary eventually reached the ventricle, and the tumor invading the septum pellucidum was also removed (Figs. 2D and 2E). Postoperatively, consciousness disturbance, fever, and meningeal irritation symptoms were prolonged but gradually recovered. Adjuvant chemoradiation therapy was initiated on the 17th postoperative day. Subsequently, the patient's clinical course was uneventful, and he received temozolomide (TMZ) maintenance therapy at our outpatient department. Local recurrence was noted at the corpus callosum 7 months postoperatively (Fig. 3), and he was treated with a gamma knife. Bevacizumab was added to the TMZ maintenance therapy after this recurrence. His condition gradually deteriorated at 15 months postoperatively, and he was subjected to terminal care.

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Fig. 1 Magnetic resonance imaging on admission at our hospital. Axial T1-weighted (A), T2-weighted (B), and T1-weighted images with contrast enhancement (C).

Fig. 2 CT scan on admission (A) and on the 8th day after symptom deterioration (B). Axial T1-weighted magnetic resonance imaging (MRI) (C) with contrast enhancement obtained on the 8th day after symptom deterioration. Axial T1-weighted MRI (D) with contrast enhancement and T2-weighted (E) postoperatively.

Fig. 3 Axial T1-weighted (A) MRI with contrast enhancement and T2-weighted (B) at the

Discussion

To the best of our knowledge, no studies have reported the rupture of cystic GBMs. Although the mechanism of cyst formation in GBM is unknown, one proposed mechanism is breakdown of the blood–brain barrier,⁴⁾ which results in extravasation of protein and water, leading to cyst formation. In the present case, we suspect that the cyst pressure elevated

due to continuous extravasation of plasma protein and water into the necrotic cyst of GBM. We must be aware that necrotic cysts may occasionally rupture in brain tumors with high proliferative ability, such as GBMs. In the present case, chemical meningitis accompanied the cystic rupture. Fortunately, this sequela was controllable in our case; however, it may lead to prognostic exacerbation.

time of recurrence.

On the other hand, the survival outcome of GBMs with cysts is reportedly better than that of GBMs without cysts. Maldaun et al.⁵⁾ have reported a series of 22 cystic GBMs and showed the tendency of better recurrence-free (7.6 vs. 4.3 months) and overall (18.7 vs. 14.4 months) survival than GBMs without cysts. They also analyzed specimens in which the relationship of the cyst wall and the brain could be analyzed and demonstrated that cystic GBMs appear to have a relatively narrow pericystic rim of glioma, with limited infiltration of the surrounding neuropil.⁵⁾ From these observations, they suggested that cystic GBMs can be treated more successfully than GBMs without cysts.⁵⁾

Another condition that causes tragedious changes in the course of GBMs may be intratumoral hemorrhage. The outcome of patients with GBMs complicated with intratumoral hemorrhage can be found in the literature. The short-term prognosis of patients with GBMs complicated with intratumoral hemorrhage is reported as approximately 22-31% mortality at 1 month, whereas 48-75% had partial or complete independence at discharge. In the long-term prognosis of intracerebral hemorrhage with GBM, the underlying malignancy prognosis is often poor at 78% mortality at 1 year.6) However, surgical removal of both the hematoma and tumor with adjuvant treatment are reportedly associated with a prolonged survival rate.7) To date, to the best of our knowledge, there is no report on prognosis of cystic GBMs complicated with cystic rupture. Nevertheless, this is a single case report, and surgical intervention may also be associated with better survival in GBMs complicated with cystic rupture. Together with the fact that cystic GBMs are associated with better outcomes, GBMs accompanying a large cyst should be treated as soon as possible.

Conclusion

We demonstrated a rare case of patient with a cystic GBM that ruptured into the cerebral ventricle. We should consider that there is a risk of rupture in cystic GBMs. Surgical intervention should be addressed as soon as possible because better prognosis is indicated in GBMs accompanying large cysts.

Conflicts of Interest Disclosure

There is no disclosure of funding as no financial support or grants supported this paper. No author has a personal or institutional financial interest in the drugs, materials, or devices described in this paper.

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