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Case Report

Spontaneous Intracerebral Hematoma in Low-Grade Glioma After 14 Years of Follow-Up

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ABSTRACT

We are reporting the case of a 53-year old woman presenting to our hospital with a hemorrhagic low-grade glioma (LGG). She was admitted to a nearby general hospital where she had presented with aphasia, right hemiplegia and change of mental status. Computer tomography (CT) images showed a left temporo-parietal hemorrhage with mass effect. She was transferred to our hospital neuro-intensive care unit where emergency craniotomy was performed. A tumor with hematoma was removed and further histopathology analysis revealed tumor progression.

We reviewed the literature reporting cases of central nervous system tumors hemorrhage and found that these types of events are exquisitely rare in adults with LGG. However these events are possible, suggesting that it should be included in the differential diagnosis of any patient presenting with intracranial hemorrhage. This case raises questions regarding the benefit of early versus late intervention for patients known to have LGG.

KEYWORDS: Intracerebral hematoma, Low-grade glioma, Surgery

INTRODUCTION

Although intratumoral hemorrhages can occur with any brain tumors, it is a very rare occurrence with low-grade glioma (LGG). In general, brain tumors are associated with intracranial hemorrhage 1-10% of the time, with an average incidence of 3.3%. In clinical series, gross hemorrhage from gliomas was seen in 1.6-10% , while the incidence of hemorrhage for all cerebral neoplasms ranged from 0.7 to 6%. Intracranial hemorrhage is commonly seen in children (1,2).

We reviewed the literature of cases with bleeding in central nervous system tumors and reported our case of hemorrhagic low-grade glioma in a 53-year-old woman presenting with right hemiplegia, aphasia and coma. Computer tomography (CT) images showed a left temporoparietal hemorrhage with mass effect.

CASE REPORT

The patient was a 53-year-old woman presenting with acute onset right hemiplegia and coma. CT scan showed a tumor in the temporo-parietal region of the brain in the left dominant hemisphere (in the insular region). Before this admission, we had followed this patient for thirteen years for a tumor in the temporo-parietal region. The initial disease presentation was a Jackson seizure. A CT with and without contrast was performed and showed the existence of a hypodense lesion in the relevant region (Figure 1). This lesion was not associated with any mass effect. Based on CT characteristics, the lesion was felt to be most consistent with a LGG. Taking into account the localization and the fact that we were dealing with a tumor in the dominant hemisphere, the decision was taken to postpone surgery and follow-up. The patient was prescribed antiepileptic treatment. The plan for regular monitoring meant primarily to perform diagnostic CT and/or magnetic resonance imaging (MRI) every 6 months for the first year, and then each

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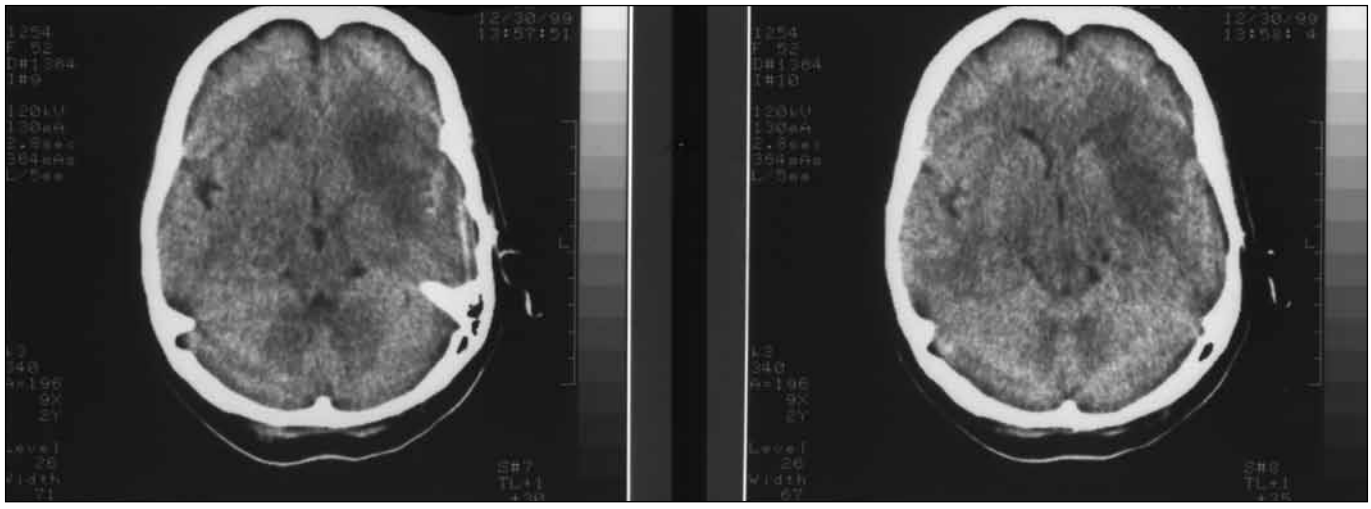

Figure 1: First CT from 1999.

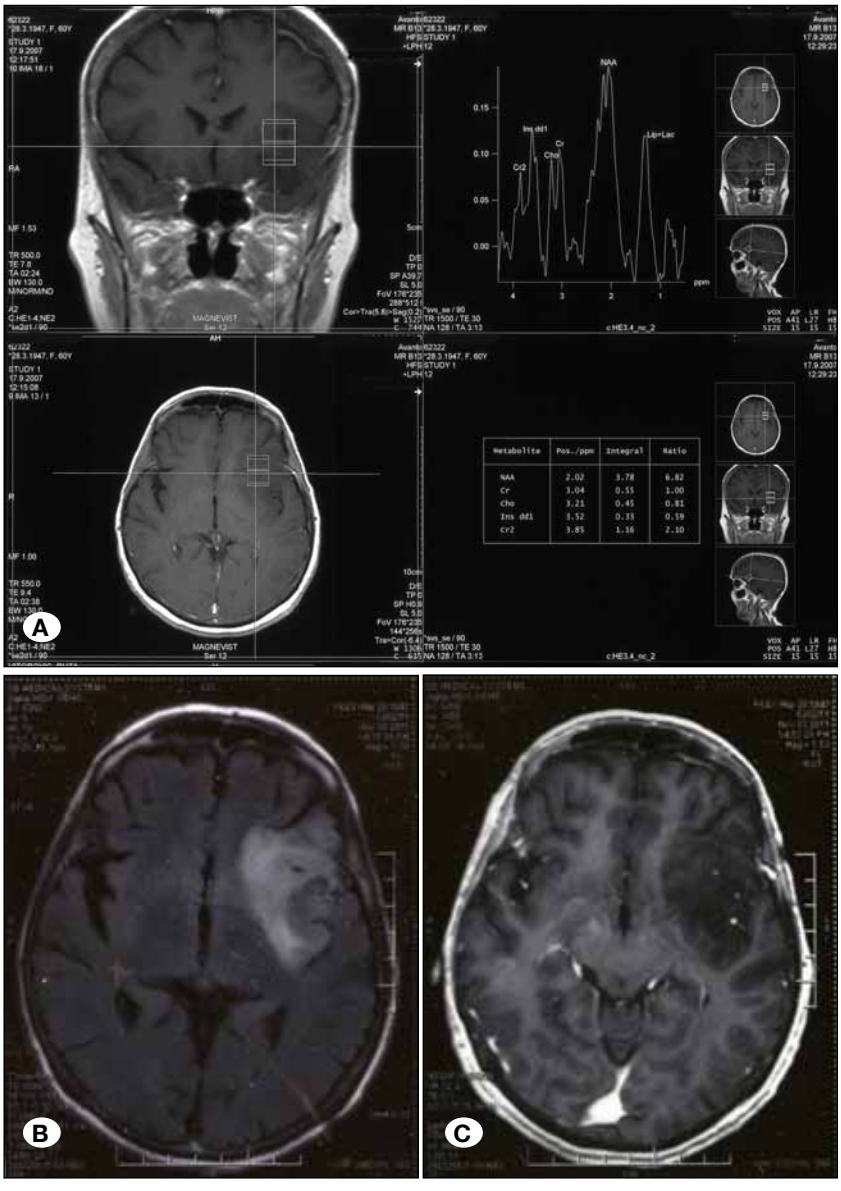

Figure 2: A) MR spectroscopy in 2007. **B)** T2 Flair, **C)** T1-W MRI in 2011 show left insular tumor.

year. Intraoperative monitoring was not available in the country at that time. A year later, the patient had a second seizure but this had no clinical consequences (normal neurological status, no subjective symptoms). A control CT with and without intravenous contrast did not show tumor progression. MRI was performed and confirmed the presumed diagnosis of LGG (Figure 2A-C). MR spectroscopy revealed reduction of NAA and elevation of Cho, myoinositol and lipid peak (Figure 2A).

The patient was started on a higher dose of antiepileptic drugs and she did well, with no seizure activity and no symptoms until October 2013, when she developed a sudden and severe headache with weakness of the right extremities and mild dysarthria after waking up in the morning. By the time of her examination in the nearest general hospital, she had developed complete right hemiplegia and aphasia. A CT scan was performed, and showed an intracerebral hematoma on the left side of the temporo-parietal part of the brain, in the same region where the LGG had previously been identified (Figure 3).

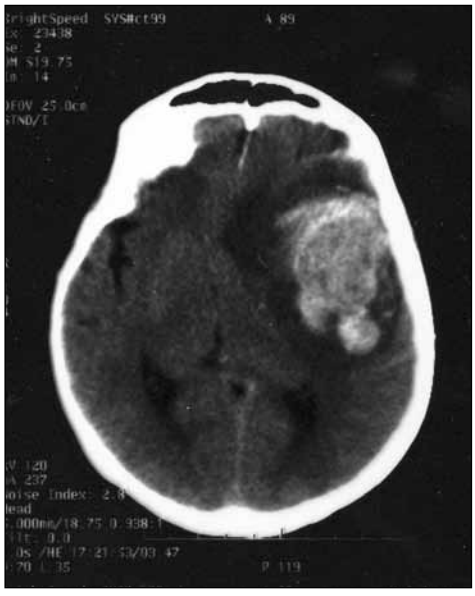

Figure 3: Preoperative CT in 2013.

She was transferred to the intensive care unit in our hospital. During transport, and shortly before admission to our hospital, there was a disturbance of consciousness leading to coma requiring artificial ventilation support. The GCS score was 7 with right hemiplegia and a discreetly asymmetrically enlarged left pupil. After preoperative preparations, emergency surgery was performed.

An intraoperative tumor was found with markedly proliferating blood vessels and with the presence of spontaneous intratumoral and peritumoral hematoma. The tumor with hematoma was removed. She was extubated on the first postoperative day and postoperative recovery was gradual. She recovered consciousness and the neurological deficit gradually decreased over the next couple of days. On the third postoperative day, the right hemiplegia changed to a barely noticeable hemiparesis and she started recovering the capacity of pronouncing certain words. She was started on early physical treatment. A control CT scan on the first post-operative day showed a small blood collection and no signs of residual tumor.

Histological examinations showed an anaplastic astrocytoma WHO grade III with the classic image of fibrillar astrocytes, without an oligodendroglial component and calcification The tissue was well vascularized and some blood vessels showed the existence of endothelial hyperplasia (Figure 4A,B). Immuno-histopathological analysis showed GFAP + + +, CD57 + + + + + p53, WT-1 and Synaptophysin + + + +, while S100, vimentin, CD10, RCC, TTF1, CK and AE1/AE3 were negative. Ki67 was 16.86%.

Based on the above history and histopthological findings, the neurooncological council made the decision to administer postoperative radiotherapy with concomitant chemotherapy.

Four months later the patient was communicative and independently mobile with full recovery of the neurological deficit

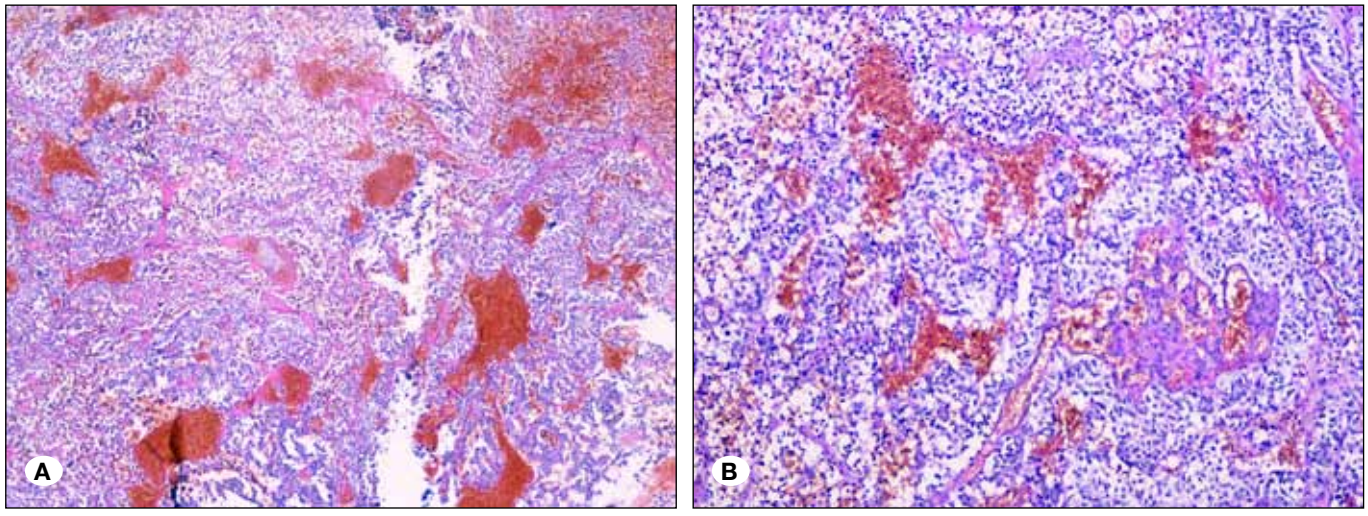

Figure 4: Histopathological findings; **A)** tumor with bleeding (HE x40); **B)** tumor with bleeding and neovascularisation (HE x100).

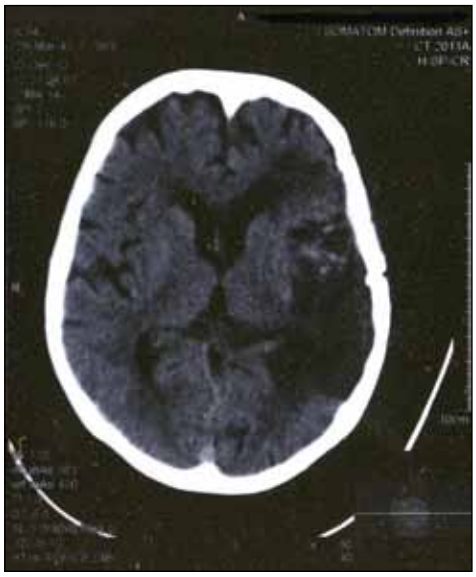

Figure 5: Postoperative CT four months after operation.

(right hemiparesis). Repeated CT scans showed an intraparenchymal defect at the site of the tumor apoplexy, and no residual tumor (Figure 5).

■ DISCUSSION

At the time of her initial presentation, it was felt that the patient had a LGG occupying part of her dominant hemisphere. Due to the tumor characteristics, as well as the fact that our patient had no neurological deficits, it was felt that the risk/benefit ratio associated with surgery was too high, and it was decided that a watch and wait approach was the most reasonable. The patient was followed for thirteen years with regular follow-up CT / MRI, which showed no signs of tumor progression and no changes in enhancement. Before the bleeding episodes, no clinical or radiographical signs of disease progression were seen. Intracerebral hematoma is an unusual clinical presentation for low-grade gliomas and it has been described in a small number of cases in adults (3,4,5). Bleeding in glioma is described mainly in tumors that have malignant properties (1).

The reasons for the emergence of intratumoral bleeding in low-grade gliomas are unclear. Probable causes of bleeding are abnormal formation of intratumoral blood vessels and endothelial proliferation within them. As the histopathological findings in our patient indicate, endothelial hyperplasia was seen in a number of intratumoral blood vessels. Abnormal and weak blood vessels are mainly associated with higher grade gliomas. This fits with the findings seen in our patient in whom dedifferentiation and progression from a low-grade glioma in an anaplastic form (WHO gr III) appears to have occurred . It is now generally accepted that the main cause of mortality in these tumors is their dedifferentiation to a higher degree of malignancy (7,8,9). Clinical studies of miR-544 in tumor tissue have shown that expression decreased significantly in secondary anaplastic astrocytoma, and in cases of spontaneous progression in malignant forms (6).

This case illustrates that the decision to delay surgery for patients with LGG should perhaps be changed, and that it may in fact be beneficial for patient to have surgery sooner rather than later.

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