Case Report

Coincidental Feeding Artery Aneurysm Presenting during Glioblastoma Surgery: A Case Report and Literature Review

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ABSTRACT

The medical literature reports an association between cerebral neoplasms and aneurysm formation. Some related aneurysms are detected during preoperative screening, whereas others are detected during or after surgery. We report a patient with de novo cerebral feeding artery aneurysm that we managed during glioblastoma surgery. We present a brief review of the literature on the coexistence of brain tumors, particularly that related to high-grade glioma and aneurysms. The literature discusses several mechanisms underlying tumor formation accompanied by aneurysm formation. Some classifications were also proposed for grouping such aneurysms. We question the necessity of the routine use of vascular imaging for patients with glioblastoma.

KEYWORDS: Feeding artery, Aneurysm, Glioblastoma, Brain tumor

■ INTRODUCTION

The hypervascularisation of brain tumors, particularly high-grade gliomas, is challenging to treat. Although rare, the literature reports an association between cerebral neoplasms and aneurysm formation (6,12). An aneurysm is expected in approximately 0.2–1.1% of patients with brain tumors (21). Some related aneurysms are detected during preoperative screening, whereas others are detected during or after surgery.

Here we report a patient with *de novo* cerebral feeding artery aneurysm that we managed during glioblastoma surgery. We present a brief review of the literature on the co-existence of brain tumors, particularly that related to high-grade glioma and aneurysms.

■ CASE REPORT

A 53-year-old woman presented to the Department of Neurosurgery, Ankara University School of Medicine. She had a 2-week history of headache and dizziness that had gradually increased. Seizure, syncope or any other symptoms were not reported. She had previously presented to the Department of Psychiatry with related symptoms. Cranial magnetic resonance imaging (MRI) revealed a 4.8×4.5×4.5 cm, poorly marginated, diffusely infiltrating necrotic lesion with peripheral contrast enhancement in the right frontotemporoparietal region (Figure 1). The mass lesion and related edema involved the capsula interna, thalamus, right cerebral peduncle and mesencephalon, suggesting a high-grade glioma. Diffusion-weighted MRI indicated a diffusion restriction in the lesion. A midline shift of 8 mm and compression of the right lateral and third ventricles warranted emergency surgery.

The patient underwent right frontotemporal craniotomy and gross total excision of the tumor was achieved. During the fine resection, a feeding artery aneurysm was observed, and it was clipped (Figure 2). The mass lesion was necrotic and highly vascularised. The patient did not exhibit a neurological deficit after surgery. Figure 3 presents the computed tomography image after surgery showing the aneurysm clip in the tumor region.

Microscopic examination of the tumor specimen revealed a poorly differentiated high-grade neoplasm with atypical

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mitotic figures, supporting the diagnosis of a World Health Organization (WHO) grade IV glioma. The patient was followed-up for 10 months. She received palliative chemoradiotherapy, but unfortunately died because of a pulmonary infection during chemotherapy.

We used the terms 'glial AND aneurysm', 'feeder aneurysm' and 'glioblastoma AND aneurysm' to conduct a systematic literature review by searching the PubMed database through September 2016, and studies based on both glioblastomas and aneurysms were included in this paper. We excluded papers published in languages other than English. The references included in each selected paper were searched as well to avoid missing any relevant study.

No significant results were obtained using the terms 'glial AND aneurysm' and 'feeder aneurysm', and searches using the term 'glioblastoma AND aneurysm' identified eight papers among 58 that included two reports on feeding artery aneurysms; one with recurrent oligodendroglioma and the other with recurrent glioblastoma, both of which were detected after chemoradiation therapy (9,24).

■ DISCUSSION

Malignant gliomas are the most common primary malignant brain tumors with an incidence of 4.13 per 100,000 (19). These neoplasms exhibit significant high cellularity, mitotic activity, necrosis, vascular fibrosis and proliferation. The incidence of coexistent primary brain tumor and aneurysm is 0.19%–4% (8,11,21,29,31). The frequencies of tumors associated with aneurysms are as follows: Meningioma (44%), glioma (38%) and pituitary adenoma (20.6%). Such association are also found in tumors, such as lymphomas, craniopharyngiomas, chordomas, epidermoid/dermoid tumors (5,17,22,26,30). Most tumor-related aneurysms are located on the internal carotid artery (26%), middle cerebral artery (26%), anterior cerebral artery (17.4%), anterior communicating artery (13%) and posterior communicating artery (4.3%)(23).
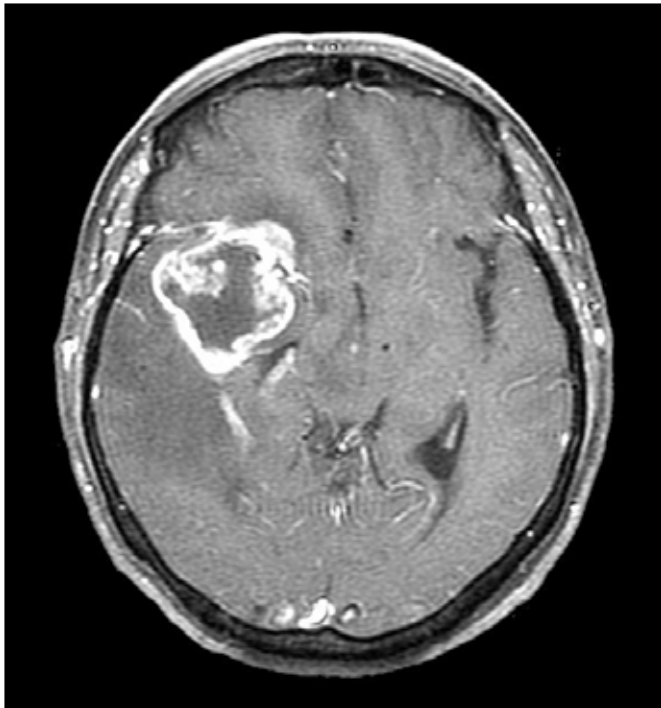
Figure 1: Preoperative T1W axial MRI scan with contrast reveals a poorly marginated, diffusely infiltrating necrotic mass lesion and midline shift.
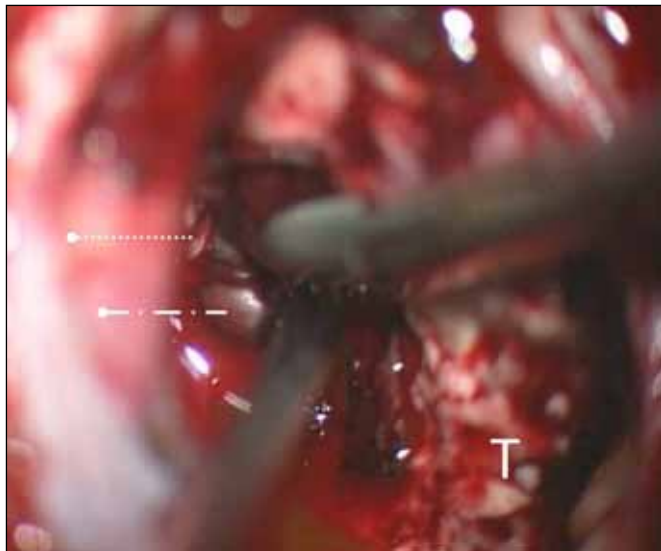
Figure 2: Glial tumor (T); feeding artery (dotted line); saccular feeding artery aneurysm (dash-dot line).

Figure 3: Postoperative axial CT scan with contrast shows the aneurysm clip in the region of the tumor.

The literature discusses several mechanisms underlying tumor formation accompanied by aneurysm formation. For example, the presence of highly fragile neoplastic vessels with irregular corkscrew-shaped lumens was suggested as the basis of the pathophysiology of aneurysms (10,14). In contrast, changes in flow dynamics and stress as well as factors, such as growth hormone, may contribute (16,20,24). Moreover, an excess secretion of growth factors, such as vascular endothelial growth factor, maintains the hypervascularity of glioblastomas (3,27). The mechanism of aneurysm formation involves the neoplastic invasion of nascent vessels and systemic coagulopathy (9). Radiation-induced vasculopathy is associated with the development of post-radiotherapy aneurysms, leading to intimal narrowing and thrombosis (1). The theory of arteriovenous shunting is supported by the presence of arterialised veins within glioblastomas to form flow-related aneurysms (18). The patient reported here was recently diagnosed with a malignant cerebral tumor, supporting the *de novo* formation of the aneurysm.

The majority of patients with tumor-associated aneurysms present with tumor-related features, and only 25% present with aneurysm-related features, such as subarachnoid haemorrhage (SAH) (21). Further, 80% of such patients present with tumor-related symptoms, and only 8% have SAH (23). These findings indicate the potential preoperative failure to detect such vascular pathologies. Moreover, a report on a unique patient with intraoperative SAH questions the existence of intra-tumor aneurysms (7).

Some classifications proposed grouping such aneurysms. A neoplastic aneurysm is defined as neoplastic cell-related emboli that may cause vessel wall invasion (2). Patients who present with an aneurysm after tumor resection may harbour a vascular injury that is classified as a pseudoaneurysm (15), and cerebral aneurysms occur after cranial irradiation (25). Another classification scheme for aneurysms associated with glioblastoma comprises aneurysms (designated UnA) that are located remotely from the tumor located on a vessel that is not related to the tumor burden, aneurysms of artery that feed the tumor (designated FsA), and false aneurysms (designated PsA) that do not possess the three layers of the vessel wall (23). This scheme emphasises that the majority of aneurysms are unrelated to the tumor. We believe that the FsA in our patient formed simultaneously inside the glial tumor, which is consistent with that in another study on an aneurysm associated with intra-tumor flow (4).

There is one report on a glioblastoma multiforme-related feeding artery aneurysm in a patient with recurrent glioblastoma and a history of irradiation, in which the authors reported routine use magnetic resonance angiography, conventional angiography or both before surgery to treat patients with gliomas (9). This strategy differs from ours because we do not routinely use any type of angiography. Therefore, we question the necessity for the routine use of vascular imaging for patients with glioblastoma.

The management of such aneurysms represents a major issue for treatment. Moreover, we believe that detecting these malformations when establishing a diagnosis is as vital as excising the tumor. The presence of an aneurysm inside a glial tumor may be overlooked because of gross bleeding during tumor excision. Therefore, examining the vascular structure of high-grade glial tumors before surgery may be safer for the surgeon and patient. Some authors suggest treating the more symptomatic lesion first (23). For example, the mortality rate is 38% for such patients whether the tumor and the aneurysm are treated separately or together (21). Generally, a simultaneous treatment of the aneurysm and glial tumor is preferred because of good outcomes (13,26,28), and prognosis remains unchanged when the tumor or aneurysms present separately or together (17).

■ CONCLUSION

Although rare, we report the second patient in the literature, presenting with glioblastoma with a feeding artery aneurysm. Published data and our experience here question whether routine vascular imaging is required before surgery to treat a high-grade glial tumor. Adopting such a strategy may help surgeons to avoid life-threatening complications, such as the rupture of an intraoperative aneurysm. Moreover, these pathologies should be excluded to reduce adverse events after surgery.

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