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Title

Hammock middle cerebral artery and delayed infarction in the lenticulostriate artery after a staged resection of giant insular glioma: A case report

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Abbreviations list: Low-Grade Glioma (LGG), Lenticulostriate Artery (LSA), Middle Cerebral Artery (MCA), Inferior Frontooccipital Fasciculus (IFOF), Direct Electrical Stimulation (DES)

Abstract*Background*

Delayed infarction in the lenticulostriate artery (LSA) area after insular glioma resection are not common and its pathophysiology remains unknown.

Case Description

A 32-year-old right-handed man with a giant insular low-grade glioma with frontal and temporal extension underwent awake craniotomy with an intentional staged surgery strategy. Preoperative radiological images demonstrated a diagonally elevated middle cerebral artery (MCA) by the temporal tumor and the a significantly compressed striatum. With intraoperative subcortical direct electrical stimulation, the resection was finalized in the temporal part of the tumor due to the semantic paraphasia induced in the temporal stem, fatigue, and loss of concentration. The immediate postoperative clinical course was uneventful. However, on postoperative day 20, he suddenly experienced right hemiparesis. Repeated images revealed infarction in the LSA area. The previously compressed striatum was then relieved and relocated to its original position in just 20 days, and the M1 segment of the MCA was remarkably downward, in which MCA resembled a hammock. Angiography confirmed the hammock-shaped MCA and significantly stretched LSA, suggesting the combination of freed striatum from the compression and the loss of temporal structure by the tumor resection as the key mechanism of the severe dislocation of the MCA and the delayed ischemia.

Conclusions

In a staged resection of giant insular glioma, attention should be paid to a possible severe dislocation of the MCA in a delayed postoperative period which may leads to LSA stretching and delayed infarction.

Highlights

1. Infarction in the lenticulostriate artery area after giant insular glioma resection is a rare presentation.
2. A staged resection, freed striatum from the compression, and the loss of temporal structure by the tumor resection may be involved in the severe dislocation of the MCA and stretched LSA.
3. Attention should be paid even in cases with uneventful immediate postoperative period.

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Introduction

The insula is the common location for low-grade gliomas (LGGs) accounting for about one-third of all LGGs.¹ Insular gliomas are still one of the most challenging tumors to manage surgically, as the insular lobe is located in the depth of the sylvian fissure and hidden by critical vascular and neural structures. These structures include the frontal and temporal operculum, precentral and postcentral gyri, middle cerebral artery (MCA), and its perforators. The lenticulostriate artery (LSA) is well known to pass through the anterior perforating substance, and the long insular artery (LIA) from the M2 segment of the MCA; these two arteries are the two main perforators at high risk of injury during resection. Furthermore, at least two language-related subcortical fiber pathways are running under the insula, namely, inferior frontooccipital fasciculus (IFOF) and the arcuate fasciculus (AF)/superior longitudinal fasciculus (SLF).² Moreover, this area plays an important role in auditory, vestibular, somatosensory, perception, taste, olfaction, motor control, and speech production.³ Nowadays, the standard of care for insular LGGs is maximal safe resection, often with intraoperative cortical and subcortical mapping by direct electrical stimulation (DES) under the awake craniotomy.⁴ The reported surgical morbidity rate following the resection of insular LGGs are ranged from 14% to 59%⁵⁻⁹ for the transient deficit and up to 20%^{5, 8, 10-13} for the permanent. Most postoperative motor deficits are caused by a deep ischemic stroke or mechanical damage in the internal capsule due to 1) direct damage to M1 and M2 segments of the MCA and their perforators including the LSA and LIA, 2) vasospasm related to the surgical manipulation around those vessels⁹, and 3) direct damage to the corticospinal tract. Those symptoms significantly impair the quality of life of patients. Considering that those surgery-related deficits are usually observed in the acute postoperative period, little attention is paid if the acute postoperative course is uneventful.

A staged surgery for the resection of giant insular gliomas, regardless of intentionally or not,

is a feasible and reasonable strategy for brain function preservation and possible neuroplasticity induction, which is a great advantage for the next surgery. However, delayed complications associated with a staged resection approach are hardly known since surgical morbidity associated with insular glioma resection is normally observed in the immediate postoperative period.

Here, we report a case of a giant insular LGG, which demonstrated delayed infarction on postoperative day 20 in the LSA area due to progressive and severe dislocation of the MCA, that is, hammock MCA. This phenomenon is unknown at all for insular glioma neurosurgeons and has great implications including its underlying mechanism and surgical management of insular LGGs, particularly when a staged resection is planned.

Case Report

History and Examination. A 32-year-old right-handed man visited a local hospital with a four-month history of memory disturbance. His magnetic resonance (MR) images demonstrated a well-defined, large hyperintense lesion on T2-weighted image in the left dominant insular lobe with frontal and temporal extension without contrast enhancement after gadolinium injection, which was well compatible with LGG (Fig. 1A-C). MR angiography revealed the diagonal M1 portion of the left MCA and the lifted M1-M2 bifurcation (Fig. 1D) due to the large temporal part of the tumor. The preoperative tumor volume was 170 mL, and the tumor was classified as giant according to Berger-Sanai classification because of its four zones of occupation⁶.

He was a consumer electronics designer but resigned 4 months before the operation personally because of the subjective deficit of logical thinking, concentration, and attention with normal findings on routine neurological examination. The Edinburgh inventory indicated that he was right handed.¹⁴ Language evaluations performed using the Western

Aphasia Battery, the DO 80 (oral denomination 80), and the Pyramids and Palm Trees Test were normal. Other preoperative neuropsychological assessments evaluating psychomotor speed/attention (Wechsler Adult Intelligence Scale, third edition; WAIS- III, Stroop test, and trail making test), working memory/attention (forward and backward digit span and the long-term reproduction of the Rey-Osterrieth complex figure test), and executive function (categorical and phonological fluency, Trail Making Test, and Stroop Test) and the Asian version of Read the Mind in the eyes test¹⁵ were also normal, except for an impaired processing speed in WAIS- III.

Operation and Pathological Findings. The patient underwent awake craniotomy with cortical and subcortical DES based on Montpellier technique¹⁶ with a biphasic current (pulse frequency, 60 Hz; single pulse phase duration, 1 msec; amplitude, 3.0 mA) applied to the cortical/subcortical DES. During the procedure, the temporal tumor was successfully removed with the subpial dissection. Insular tumor was partially removed with the transopercular approach and continuous subcortical DES. The resection was intentionally finalized according to the patient's fatigue, poor concentration, and the functional boundary, where the semantic paraphasia was induced by the DES to the left IFOF in the temporal stem (awake period, 98 mins). Postoperative MR images on the next day of surgery confirmed the total resection of the temporal tumor but large residue in the insular lobe (Fig. 1E) with the extent of resection of 45%. The left striatum was still markedly displaced medially by the insular tumor the same as preoperatively at this point. The pathological diagnosis was oligodendroglioma, IDH-mutant with 1p/19q codeletion.

Postoperative Course. The acute postoperative course was uneventful. Detailed objective neuropsychological assessment on postoperative day 10 revealed no motor, sensory, or

cognitive dysfunction except for a slight delay in the executive function in the trail making test, and he was discharged on that day. However, 20 days after the operation, he suddenly experienced the right hemiparesis, and repeated MR images revealed multiple infarctions in the LSA area (Fig. 1F). Notably, the displaced striatum then relocated to its original position in just 20 days, and the left M1 portion was significantly in downward direction (Fig. 1G), which made the MCA in a hammock shape (Fig. 1H). The resection cavity in the middle temporal fossa was totally occupied by the herniated insular tumor. Conventional angiography confirmed the hammock-shaped MCA without other vascular lesions (Fig. 1H), which suggested that the gradually and progressively stretched LSA led to the delayed infarction.

Discussion

Major complications associated with resection of insular glioma are 1) ischemic stroke caused by direct damage to LSA or LIA, 2) direct damage to the corticospinal tract, and 3) subcortical fiber pathways including IFOF and AF/SLF. These lead to motor and/or language deficits usually in the immediate postoperative phase, and the gradual recovery of those neurofunction is expected in some patients, especially those who underwent functional cortical and subcortical mapping with awake craniotomy and the preservation of those white matter pathways.¹⁷

Two main approaches are available for resection of insular glioma: transopercular approach (TOA)² and transsylvian approach (TSA).¹⁸ The cadaveric comparative study between TOA and TSA by Benet¹⁹ revealed better insular exposure and surgical freedom in TOA than TSA. It is also suggested that under the removal of the operculum with cortical and subcortical DES mapping, the TOA may have an advantage even in zone II and III tumors, where the facial motor, somatosensory, and language functions are at risk of damage. Because the TSA

often requires sacrifice of some superficial sylvian veins²⁰ and retraction of the operculum, we routinely perform TOA with cortical and subcortical mapping under awake craniotomy regardless of whether pure insular gliomas or giant gliomas. In this case, first, the resection of the temporal tumor was performed according to the posterior functional boundary, where the semantic paraphasia was induced in the posterior margin of the tumor. Second, the frontal operculum was removed without any kinds of paraphasia. However, in the depth of the limen insula, the patient again demonstrated semantic paraphasia and we confirmed that the resection was very close to the main bundle of the IFOF. Since then, the concentration status of the patient became unsatisfactory, and we decided to stop the surgical manipulation, considering intentional staged resection in the near future. The postoperative course was uneventful with no new functional deficits, and MR images on the next day of surgery revealed a resection rate of 45%. However, as described above, on postoperative day 20, the patient demonstrated a sudden onset of right hemiparesis due to the hammock MCA and the remarkably stretched LSA which resulted in ischemic stroke.

Although the exact mechanism of the LSA infarction was not totally revealed, the authors speculate that the combination of 1) the gradual but progressive relocation of the displaced striatum to its original position and 2) the total removal of the temporal tumor, which was a necessary structural support for the insular tumor, were essential for the severe dislocation of the MCA and ischemic pathophysiology in this patient. Angiography revealed a significantly stretched LSA but no other vascular abnormalities. This hypothesis and angiogram findings also support the clinical time course, particularly the delayed onset of ischemia. Normally, no special attention is paid if no postoperative deficit is observed in the immediate postoperative period even in cases with large residues. Therefore, all the neurosurgeons who take care of insular gliomas must recognize this rare but critical complication, especially when a staged resection is applied.

A staged resection strategy with incomplete resection for the prevention of permanent deficit in insular glioma surgery⁴ is feasible and reasonable with the expectation of neuroplasticity induction.²¹ The next clinical question is how to avoid this delayed ischemia. As stated, the possible mechanism include partial resection of a giant insular glioma in a staged surgery (intentional or unintentional) and a relief of the compressed striatum, which leads to an anatomical shift, pushing the MCA downward and stretching the LSA. We hypothesize that the loss of physical support by the preceding temporal tumor resection also plays another key role in this rare but critical complication. Therefore, in giant insular tumors, even when a partial resection under a staged surgery policy is performed and no neurological deficit is observed in the immediate postoperative period, attention should be kept for early detection of a possible MCA dislocation. If the hammock MCA is detected, the early surgical intervention for the residual tumor should be considered for the prevention of the stretching of the LSA and ischemia.

Conclusions

The present case is rare but provides important information on delayed ischemic stroke in LSA after the resection of the temporal tumor of a giant insular LGG, which was caused by the downward dislocated MCA and the remarkably stretched LSA. This case also provides meaningful clinical implications, suggesting that in a staged resection for giant insular tumors, attention should be paid for early detection of the hammock MCA translocation.

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Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Figure Legends

Figure 1. Preoperative magnetic resonance (MR) images (A-C). MR angiogram (D) demonstrates the giant insular low-grade glioma with temporal and frontal extension. Note that the left striatum is remarkably compressed medially by the tumor. The left middle cerebral artery (MCA) is diagonal because of the temporal part of the tumor. The MR image on the postoperative day 1 (E) shows the total resection of the temporal part but with large amounts of residues in the insular part. The MR images on postoperative day 20 (F, G) reveal infarction in the LSA area. Note that the freed striatum, in combination with the removal of the temporal tumor, significantly pushes the MCA downward. Angiogram (H) showing the hammock MCA and the significantly stretched LSA, which leads to the ischemia in the LSA.

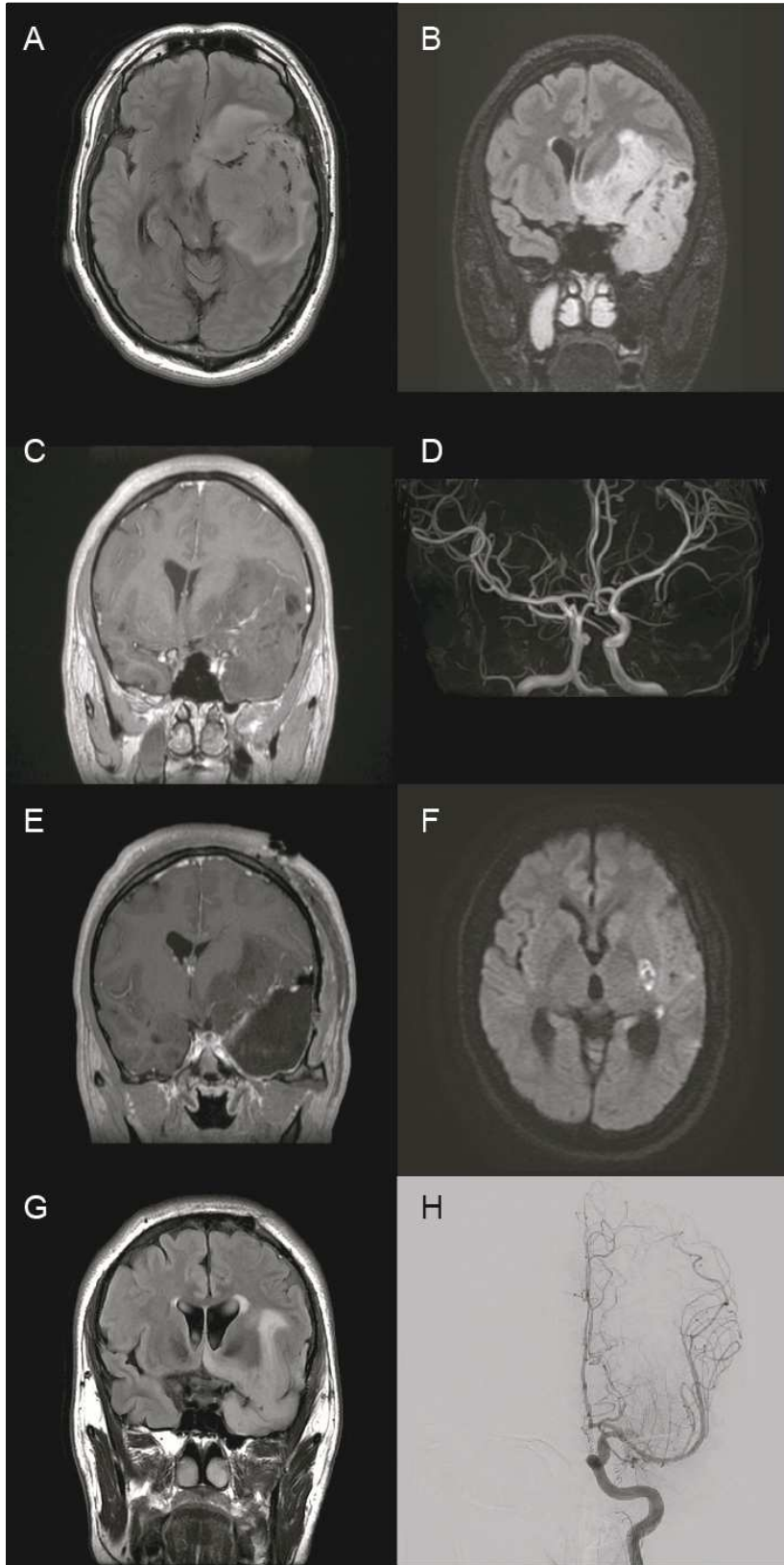
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