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Case Report

Combination of asleep and awake craniotomy as a novel strategy for resection in patients with butterfly glioblastoma: Two case reports

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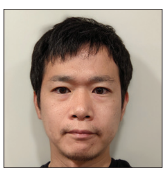

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ABSTRACT

Background: Several studies have reported that gross total resection contributes to improved prognosis in patients with butterfly glioblastoma (bGBM). However, it sometimes damages the corpus callosum and cingulate gyrus, leading to severe neurological complications.

Case Description: We report two cases of bGBM that was safely and maximally resected using brief and exact awake mapping after general anesthesia. Two patients had butterfly tumors in both the frontal lobes and the genu of the corpus callosum. Tumor resection was first performed on the nondominant side under general anesthesia to shorten the resection time and maintain patient concentration during awake surgery. After that, awake surgery was performed for the lesions in the dominant frontal lobe and genu of the corpus callosum. Tumor resection was performed through minimal cortical incisions in both frontal lobes. Postoperative magnetic resonance imaging showed gross total resection, and the patients had no chronic neurological sequelae, such as akinetic mutism and abulia.

Conclusion: bGBM could be safely and maximally resected by a combination of asleep and brief awake resection, which enabled patients to maintain their attention to the task without fatigue, somnolence, or decreased attention. The bilateral approach from a small corticotomy can avoid extensive damage to the cingulate gyrus.

Keywords: Awake surgery, Butterfly glioblastoma, Maximal safe resection

INTRODUCTION

Butterfly glioblastoma (bGBM), named after its characteristic imaging findings, is a bilateral frontal lesion that grows bidirectionally from the corpus callosum or progresses to the contralateral cerebral hemisphere through the corpus callosum. The previous studies have reported that the median survival of patients with bGBM is 3–6 months,[3,5,6,9] showing a poorer prognosis than nonbutterfly glioblastoma (non-bGBM).[3]

Several studies have reported that a greater extent of resection under general anesthesia contributes to the improvement of prognosis in patients with bGBM.[1,3,5-7] In addition, few studies have reported the efficacy of awake surgery for bGBM.[2,4] According to the previous studies, confusion (21.0–33.3% of all bGBM patients) was the second most common clinical presentation of bGBM following headaches (30.6–48.7%).[1,6] Therefore, if we choose awake brain surgery for the treatment of bGBM, it should be noted that decreased attention due to prolonged awake brain surgery makes it difficult to continue exact mapping, especially for confused patients. Here, we report two cases of bGBM that could be safely and maximally resected by brief and exact awake mapping along with a literature review.

CASE DESCRIPTION

A combination of asleep and awake resection was performed to shorten the resection time and maintain patient concentration during awake surgery. Tumor resection was performed through minimal cortical incisions in both frontal lobes to avoid damaging the cingulate gyrus. After the tumor on the nondominant side was completely resected under general anesthesia, the patient was awakened from general anesthesia. Then, brain mapping on the dominant frontal lobe was performed by bipolar 4 mA electrostimulation based on the guidelines for awake craniotomy.[8] Counting, visual naming, and auditory comprehension tests were also conducted. The tumor on the dominant side was then resected carefully while the speech therapist talked with the patient. Further operative manipulation was discontinued when speech responsiveness was reduced, even if only slightly. Both cases illustrated below were considered to be left hemisphere dominant for language, based on the fact that they and all their family were right-handed.

ILLUSTRATIVE CASES

Case 1

A 62-year-old woman developed confusion and was transported to a nearby hospital through ambulance. Occasional incoherent speech and behavior were observed from 2 days before her presentation. Computed tomography and magnetic resonance imaging revealed a hemorrhage and butterfly lesion with contrast enhancement across both frontal lobes. She was diagnosed with intratumoral hemorrhage and was transferred to our hospital 10 days after the initial diagnosis. On admission, the patient was conscious with no motor paralysis or aphasia. Her Standard Language Test of Aphasia (SLTA) score was a normal value of 226/232. Her Mini-Mental State Examination (MMSE) score was 27/30, and her Raven's Colored Progressive Matrices (RCPM) score was 25/36, slightly below the average score for individuals in their sixties. On admission, MRI showed a space-occupying lesion, where a hemorrhagic lesion and a contrast-enhanced lesion were mixed in both frontal lobes and the genu of the corpus callosum [Figure 1A]. The maximum lesion diameter was 52 mm.

Tumor resection with bilateral frontal craniotomy in normal supine position was performed using propofol under general anesthesia 18 days after the initial diagnosis. First, an approximately 2 cm incision was made in the right frontal lobe cortex, which revealed a tumor capsule with blood clots inside. The lesion was removed with a high-power SONOPET ultrasound aspirator and the gliotic border outside the tumor was dissected using a low-power SONOPET. Two hours after the start of craniotomy, the right tumor was completely resected, except for the lesion of the corpus callosum, and the patient was awakened from general anesthesia. Brain mapping of the left frontal lobe was performed using bipolar 4 mA electrostimulation, and the patient did not show any neurological symptoms. Corticotomy was performed on the left frontal lobe and the tumor with blood clots on the left side was resected carefully while the speech therapist talked with the patient. Deep-seated tumors on both sides were also resected by entering from the left and right sides of the falx cerebri until the anterior horn of each lateral ventricle was opened [Figure 1B, e]. The patient had few complaints of headache or sickness, even after ventricular opening, and responded to conversation fluently during awake surgery. The left and right pericallosal arteries were found in the deep area and the tumor that appeared to adhere to the artery was carefully dissected. Finally, the resection cavity was examined endoscopically, including the corpus callosal margin and pericallosal artery [Figure 1B, f]. The bilateral tumor was completely resected through minimal cortical incisions in both frontal lobes [Figure 1B, g]. The operative time for microsurgical resection in the awake state was 190 min. Postoperative MRI showed gross total removal of the tumor, including the lesion in the genu of the corpus callosum, and preservation of both pericallosal arteries [Figure 1C, h-i].

The patient showed no paralysis or aphasia postoperatively, with an SLTA score of 230/232. In addition, 1 week after the surgery, her cognitive function improved with an MMSE score of 27/30 and RCPM score of 33/36. Final pathology revealed isocitrate dehydrogenase (IDH) wild-type glioblastoma without a methylated MGMT promoter. She had received postoperative local radiotherapy with concomitant temozolomide followed by 12 cycles of adjuvant temozolomide with a KPS of 70. At the 1-year follow-up, MRI revealed no obvious recurrent lesions [Figure 1C, j-k]. She had a favorable progression-free survival of 20 months and overall survival of 24 months and is still alive.

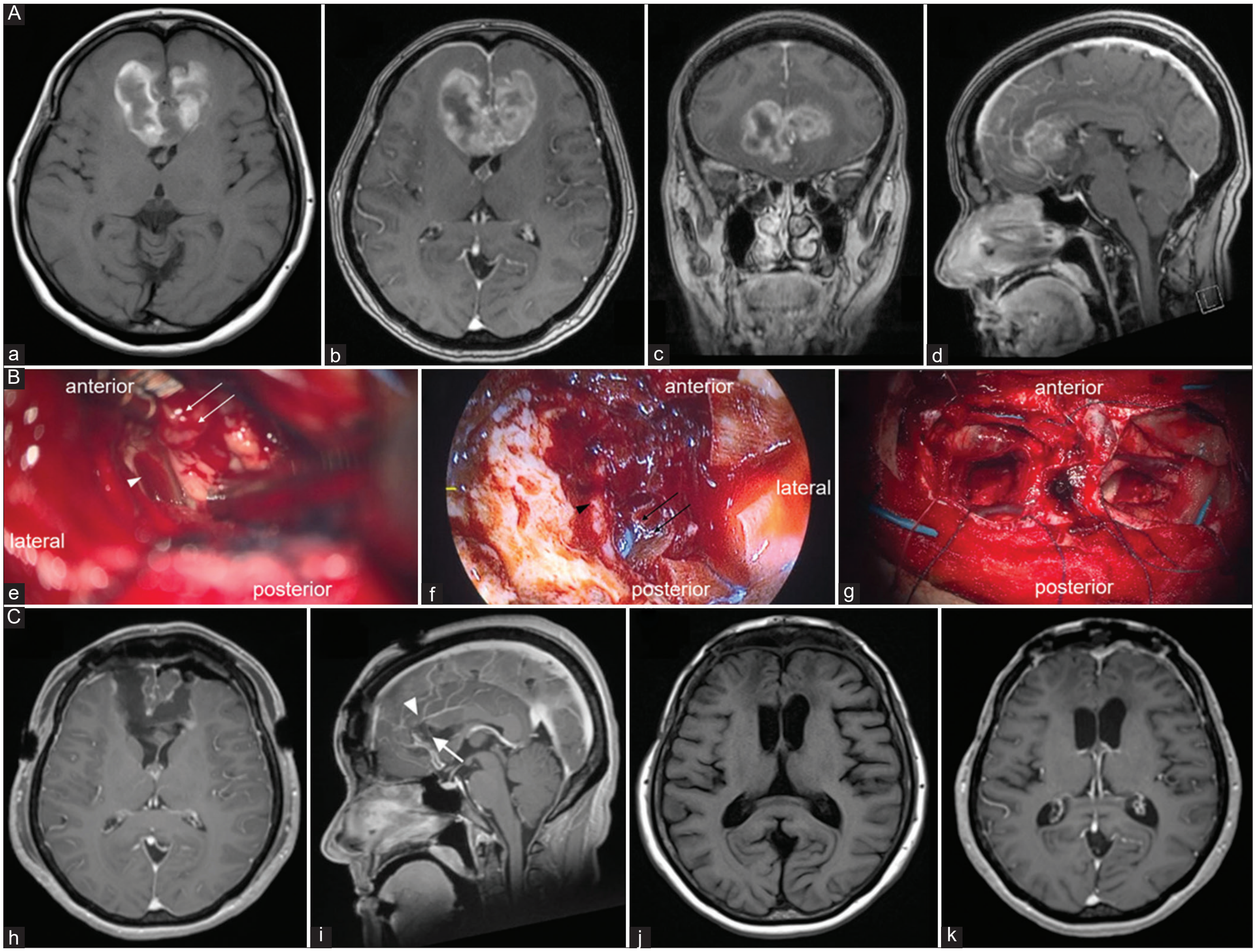

Figure 1: MRI imaging findings and intraoperative photographs in Case 1. (A) Preoperative MRI imaging including T1-weighted imaging (a; axial) and contrast-enhanced T1-weighted imaging (b; axial, c; coronal, and d; sagittal) showed a space-occupying lesion where a hemorrhagic lesion and a contrast-enhanced lesion were mixed in both frontal lobes and the genu of the corpus callosum. The maximum diameter of the lesion was 52 mm. (B) Intraoperative photographs are shown. e; Deep-seated tumor on the left side (white arrow) was resected by entering from the left side of the falx cerebri until the left anterior horn (white arrowhead) was opened. (f) The resection cavity was examined with an endoscope, inserted through the right side of the falx cerebri. The right pericallosal artery (black arrowhead) and the cavity after removal of the tumor in the corpus callosum (black arrow) are shown in this figure. (g) The bilateral tumor was totally resected through the minimal cortical incisions of both frontal lobes. (C) Postoperative MRI imaging at 1 month (h and i) and 1 year (j and k) after the surgery are shown. At 1 month, contrast-enhanced MRI (h) axial and (i) sagittal showed gross total removal of the tumor, including the lesion in the genu of corpus callosum (white arrow). Both pericallosal arteries (white arrowhead) are preserved. At 1 year, MRI (j) T1-weighted imaging and (k) contrast-enhanced T1-weighted imaging show no obvious recurrent lesion.

Case 2

A 45-year-old man noticed a pulsatile headache with a 2-month history and consulted a doctor. MRI showed a space-occupying lesion in both frontal lobes. The patient was referred to our hospital and admitted for surgery. On admission, he was conscious with no motor paralysis, aphasia, and cognitive dysfunction. His SLTA, MMSE, and RCPM scores were 231/232, 30/30, and 35/36, respectively. Preoperative MRI showed a space-occupying lesion with a mixture of hemorrhagic and contrast-enhanced lesions in the medial aspect of both the frontal lobes and corpus callosum [Figure 2A]. The maximum lesion diameter was 60 mm.

Tumor resection was performed through bilateral frontal craniotomy and minimal corticotomy. The tumor was yellowish and well-demarcated, with cysts and hemorrhage inside. The tumor on the right nondominant side was resected and the anterior horn of the lateral ventricle was opened in the deep area. Next, the left frontal lobe tumor was debulked as much as possible under general anesthesia. The falx cerebri was incised to preserve the inferior sagittal sinus

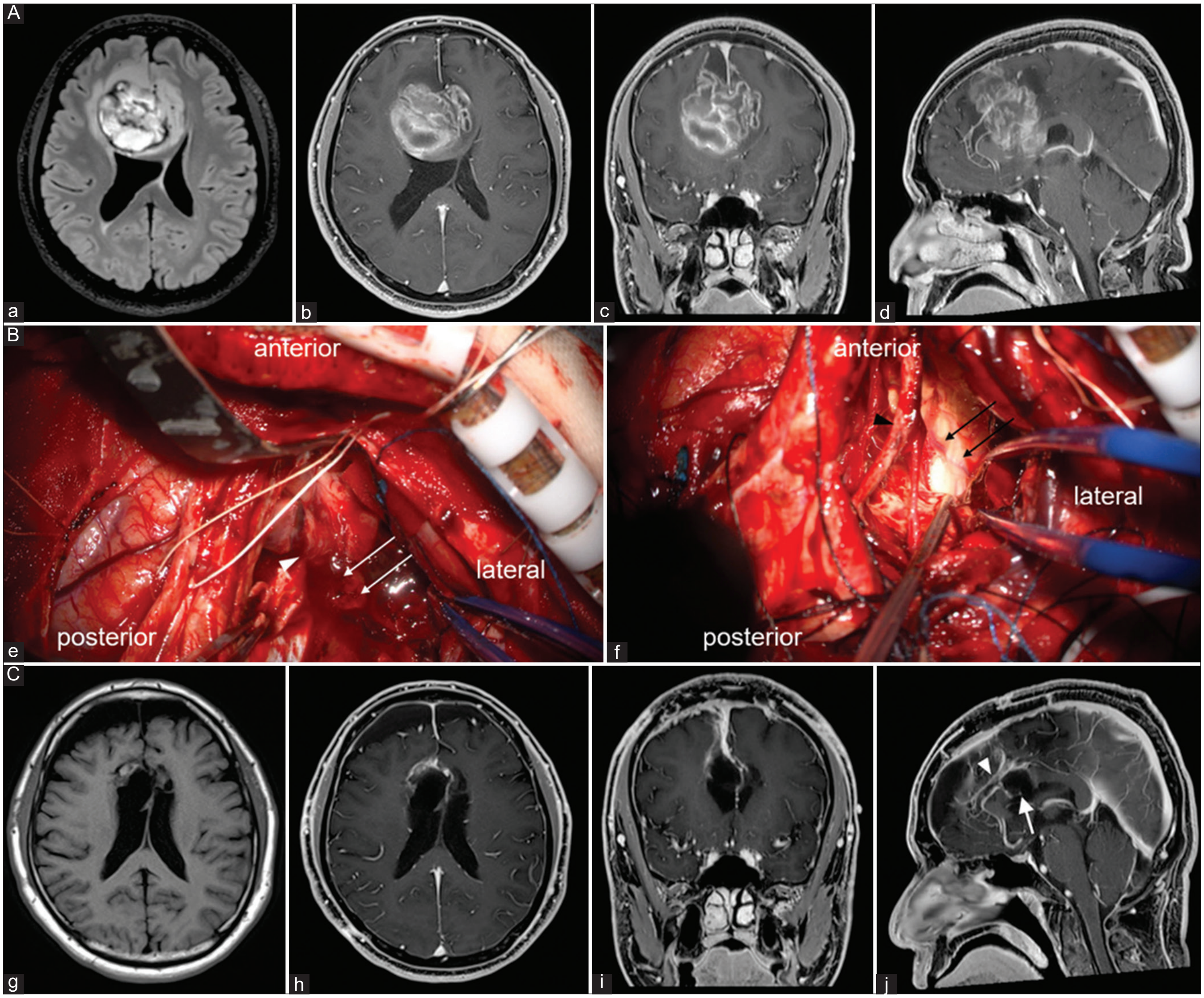

Figure 2: MRI imaging findings and intraoperative photographs in Case 2. (A) Preoperative MRI imaging including FLAIR imaging (a; axial) and contrast-enhanced T1-weighted imaging (b; axial, c; coronal, and d; sagittal) showed a space-occupying lesion with a mixture of a hemorrhagic lesion and a contrast-enhanced lesion in the medial aspect of the frontal lobes to the cingulate gyrus and corpus callosum. The maximum diameter of the lesion was 60 mm. (B) Intraoperative photographs are shown. (e) The right frontal lobe tumor was resected, and the left frontal lobe tumor was debulked as much as possible under general anesthesia. The falx cerebri was incised to preserve the inferior sagittal sinus (white arrowhead). This manipulation enabled us to resect the left-sided tumor from the right side of the falx cerebri. The patient was awakened from anesthesia with the residual tumor in the deep corpus callosum (white arrow). (f) The residual tumor in the deep corpus callosum was resected and the anterior horn of the lateral ventricle (black arrow) was opened. The right pericallosal artery (black arrowhead) and its branch vessel penetrating the tumor were carefully dissected and preserved. (C) Postoperative MRI 1 month after the surgery is shown. T1-weighted imaging (g; axial) and contrast-enhanced T1-weighted imaging (h; axial, i; coronal, and j; sagittal) showed gross total removal of the tumor, including the lesion in the anterior part of corpus callosum (white arrow). The pericallosal and callosomarginal arteries (white arrowhead) are preserved.

and the tumor was resected from the right side of the falx cerebri [Figure 2B, e]. The patient was awakened 4 h after the start of craniotomy and he underwent brain mapping as reported in Case 1. He had no neurological deficits by electrostimulation, and the residual tumor in the deep corpus callosum and the medial aspect of the left frontal lobe was carefully dissected. Simultaneously, the patient moved the limbs, counted numbers, and answered verbal questions. The tumor adhered to the left and right pericallosal arteries and their branch vessels were carefully resected using SONOPET [Figure. 2B, f]. Intraoperative MRI revealed a residual tumor in the genu of the corpus callosum, 160 min from the start of awake surgery, which was additionally resected after confirmation of stable speech responsiveness using electrical stimulation. Further operative manipulation was discontinued because the speech responsiveness was slightly reduced after resection of the dorsal part of the tumor in the genu of the corpus callosum. The operative time for microsurgical resection in the awake state was 150 min. Postoperative MRI revealed gross total removal of the tumor, including the lesion in the anterior part of the corpus callosum [Figure 2C].

One day after surgery, he presented with transient intellectual dysfunction with an MMSE score of 29/30, RCPM score of 32/36, and Frontal Assessment Battery (FAB) score of 12/18. However, 3 weeks after the surgery, his cognitive function improved, with an MMSE score of 30/30, RCPM score of 36/36, and FAB score of 16/18. The SLTA score measured at 3 weeks was 232/232. The final pathology revealed IDH wild-type glioblastoma without MGMT promoter methylation. Until now, he has received postoperative local radiotherapy with concomitant temozolomide.

DISCUSSION

bGBM involves the corpus callosum and cingulate gyrus in the bilateral frontal lobe and surgical resection has the risk of causing akinetic mutism and abulia.[2] Moreover, the risk of neurological complications may be higher in bGBM than in non-bGBM patients. The risk of severe neurological complications such as motor deficits and speech and language disturbance with resection is reported to be higher than with biopsy.[4] Therefore, biopsy only or partial resection followed by postoperative chemoradiotherapy has been relatively common treatment options for bGBM.[10] Because of this background, the median survival of patients with bGBM is limited to approximately 3–6 months, according to the previous studies,[3,5,6] showing a poorer prognosis than non-bGBM patients.[3] Meanwhile, several recent studies have reported that a greater extent of resection contributes to the improvement of life prognosis in patients with bGBM, without impairing QOL.[1,3,5-7] Chaichana *et al.*[3] compared the survival rates between the surgical and biopsy groups after matching for age, physical status, tumor size, and subsequent therapies. The survival rates at 6 months in the biopsy and resection groups were 12 and 60%, respectively, and those at 12 months were 0 and 12%, respectively. Chaichana *et al.*[3] also reported that survival is prolonged when the extent of resection exceeds 65%, but some recommend a resection extent of ≥86%[6] and ≥98%[2,9].

In light of these reports, the challenges of maximal safe resection of bGBM should be addressed. However, it is controversial whether bGBM should be resected under general anesthesia or in an awake state. According to the previous reports of surgical resection under general anesthesia, the incidence of postoperative neurological symptoms is 16–18%.[1,3] In contrast, Burks *et al.*[2] performed awake surgery by paying attention to the preservation of nerve fiber bundles in the cingulum and reported that no permanent neurological deficits, including akinetic mutism and abulia, were observed. They determined the anatomical localization of the cingulum bundle entirely by awake monitoring with attention tasks, that is, when the patient struggled with the attention tasks, they tried to redirect operative view laterally to avoid the cingulum bundle. As with the findings of Burks *et al.*, we consider that awake surgery is advantageous in attention monitoring, leading to safe and maximal resection while preserving important nerve fiber bundles in the corpus callosum and cingulate gyrus.

The problems in performing awake surgery for bGBM may include the relatively long operative time due to bilateral and extensive lesions and the complexity of the surgical operation due to the involvement of the corpus callosum and the anterior cerebral artery. Fatigue, somnolence, and decreased attention due to prolonged awake surgery make it difficult to continue the task, especially in older adults and patients with higher brain dysfunction before surgery. In addition, the effect of prolonged compression of the frontal lobe by the retractor and the effect of frequent electrical stimulation during awake surgery, which may lead to decreased patient attention, should also be minimized. Given the above mentioned, in these cases, we first resected the tumor of the nondominant right frontal lobe under general anesthesia followed by resection of the tumor of the left dominant frontal lobe and the genu of the corpus callosum in a brief awake state. Intraoperative electrical stimulation was minimized by monitoring free speech during surgery. When the surgical operation extended to the corpus callosum or cingulate bundle, the operation was interrupted or discontinued if speech responsiveness was reduced even slightly, as illustrated in Case 2.

To the best of our knowledge, this is the first case reports of bGBM that could be safely and maximally resected by combination of asleep and awake craniotomy. Further studies with larger population size are required.

CONCLUSION

The bGBM with corpus callosum involvement was safely and maximally resected by a combination of asleep and brief awake resection, which enabled patients to maintain their attention to the task during awake surgery. Tumor resection through minimal bilateral cortical incisions might have contributed to preserving the anatomical connectivity around the corpus callosum and cingulate gyrus in our cases.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

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