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Significance of novel subcortical low intensity score on transient neurological events after revascularization surgery for moyamoya disease

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A R T I C L E I N F O

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A B S T R A C T

Objectives: Transient neurological events (TNEs) are frequently observed after revascularization surgery for moyamoya disease (MMD). Recently, two signs on fluid-attenuated inversion recovery magnetic resonance images, a cortical hyperintensity belt (CHB) sign possibly reflecting vasogenic edema and a transient subcortical low intensity (SCLI) sign possibly reflecting cytotoxic edema, were reported associated with TNEs. The purpose of this study was to create a SCLI score and to investigate the significance of the score in TNEs.
Patients and methods: The authors retrospectively analyzed 18 cerebral hemispheres in 16 consecutive patients with revascularization surgery for MMD. The SCLI sign was defined as a transient SCLI in surgically treated hemispheres, and blindly graded as the SCLI score (0–4) based on the extent. The relationships among SCLI, CHB signs and TNEs were evaluated.
Results: Postoperative TNEs, SCLI and CHB signs were detected in 8 (44.4%), 9 (50.0%) and 12 (66.7%) hemispheres, respectively. Patients with SCLI and CHB signs had a significantly higher TNE occurrence rate than those without these signs ($p = 0.015$, and $p = 0.013$, respectively). Patients with TNEs showed significantly higher SCLI scores than those without TNEs ($p = 0.009$), while the difference of CHB scores did not reach significance between patients with and without TNEs. For the occurrence of postoperative TNEs, SCLI score with a cut-off value of 1.0 resulted in a specificity of 80.0% and a sensitivity of 87.5%.
Conclusion: The novel SCLI score may be useful for diagnosing TNEs after revascularization surgery for MMD, although both vasogenic and cytotoxic edema may be involved in postoperative TNEs.

1. Introduction

Moyamoya disease (MMD) is a chronic, occlusive cerebrovascular disease of unknown etiology characterized by steno-occlusive changes at the terminal portion of the internal carotid artery and an abnormal vascular network at the base of the brain [1]. Revascularization surgery is widely used as the treatment of choice for symptomatic MMD to decrease the risk of future stroke [2]. However, transient neurological events (TNEs) are relatively commonly observed after revascularization surgery for MMD [2–9]. Recently, a cortical hyperintensity belt (CHB) sign, possibly reflecting vasogenic edema, was reported as a predictor of TNEs after bypass surgery for MMD [5]. The authors also reported a transient subcortical low intensity (SCLI) sign on fluid-attenuated inversion recovery (FLAIR) magnetic resonance (MR) imaging, possibly reflecting cytotoxic edema, in a patient with MMD in association with postoperative TNEs [10]. However, the significance of SCLI signs remains unknown. In the present study, thus, we proposed a novel SCLI score and investigated the diagnostic significance in postoperative TNEs compared with previously reported CHB scores [5].

2. Patients and methods

2.1. Patient selection

All study protocols were approved by the institutional review board at Mie University Graduate School of Medicine. This was a retrospective study and included 18 cerebral hemispheres in 16 consecutive cases with MMD (mean age, 40.7 years; age range, 6–71 years; female, 11 hemispheres in 10 cases) who underwent revascularization surgery at Mie University Hospital between August 2009 and September 2016. Patient demographic data were listed in Table 1.

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Table 1
Characteristics of patients with moyamoya disease in the study.

Age at surgery (years)	
Mean ± standard deviation	40.7 ± 17.7
Range	6–71
Sex, number of patient (%)	
Male	6 (37.5)
Female	10 (62.5)
Operative side, number of hemisphere (%)	
Right	8 (44.4)
Left	10 (55.6)
Events before surgery, number of hemisphere (%)	
Ischemia	13 (72.2)
Hemorrhage	3 (16.6)
Other	2 (11.1)

2.2. Surgical procedures

Our indications of revascularization surgery for MMD were as follows: the presence of minor ischemic or hemorrhagic stroke and/or transient ischemic attack, apparent hemodynamic compromise on single-photon emission computed tomography (SPECT) defined with the previous report [11], independent activity of daily living (modified Rankin Scale scores of 0–2), and the absence of major cerebral infarction and/or massive intracerebral hemorrhage.

All surgical procedures included both direct anastomosis of the superficial temporal artery (STA) to the middle cerebral artery (MCA) and indirect bypass of encephalosynangiosis. The main branch of the STA was anastomosed to the cortical segment of the suprasylvian MCA (M4 segment) in an end-to-side fashion. If appropriate, another STA branch was anastomosed to the cortical branch of the infrasylvian part of the M4 segment. We decided whether to perform single or double STA-MCA bypasses according to the preoperative angiographic findings of the STA development or intraoperative findings of the MCA diameter being suited for direct anastomosis. In all hemispheres, indirect procedures using temporal muscle, dura mater, and pericranial flaps were performed combined with direct procedures.

2.3. Patient evaluations

Before surgery, all patients underwent cerebral computed tomography, FLAIR, diffusion-weighted MR imaging, MR angiography, with either a 1.5-T (Achieva, Philips) or 3.0-T (Ingenia, Philips) scanner, and conventional cerebral angiography, as well as N-isopropyl [^{123}I]-p-iodoamphetamine SPECT (^{123}I-IMP-SPECT) with and without acetazolamide challenge to measure cerebral blood flow and cerebrovascular reserve in the affected hemisphere. These examinations, except for cerebral angiography and acetazolamide loading SPECT, were also performed routinely during postoperative days 1–7, even if the patients did not develop TNEs. In addition, in cases with TNEs, MR imaging, MR angiography and resting ^{123}I-IMP-SPECT were performed on the day or within a couple of days after TNE occurrence. If MR imaging detected abnormal findings, follow-up MR imaging was repeated every one to 4 weeks according to the preference of neurosurgeons in charge until the abnormality disappeared.

2.3.1. SCLI sign and CHB sign in MR imaging

The criteria for identifying the SCLI sign were as follows: 1) presence of a low intensity signal on FLAIR images within the subcortex of the MCA territory of the surgically treated hemisphere; 2) the signal intensity ratio < 0.90 of the low intense subcortical white matter to the subcortical white matter in the contralateral side; and 3) disappearance of the low intensity signal in the chronic period (3–6 months after the operation). Examples of CHB and SCLI signs are shown in Fig. 1.

The CHB sign and score were identified as previously described [5]. All slices of the axial FLAIR images were reviewed to determine the extent of these signs according to the previous report [5]. The extent of SCLI sign was also defined according to the CHB scoring (Fig. 2). Briefly, the central sulcus represented the division of the MCA territory between anterior and posterior parts in all slices on axial FLAIR images. In each part, the extent of the SCLI sign was scored as 0 (not visible), 1 (limited to less than the half of the part), or 2 (extending over the half of the part). The SCLI and CHB scores were the sum of each part score (minimum = 0 and maximum = 4), respectively. These scores were determined through the consensus by 2 board-certified neurosurgeons (R.Y. and N.T.), who were blinded to neurological conditions and outcome of each case.

2.3.2. Neurological evaluation

We retrospectively evaluated patients' medical records and defined TNEs according to the previous reports as follows [3,5]: 1) reversible neurological deficits observed objectively (e.g., motor weakness, and aphasia); 2) reversible neurological deficits recognized subjectively and reported by the patients (e.g., numbness of the extremities); 3) no sign of acute cerebral infarction on neuroimages; and 4) no sign of acute intracranial hemorrhage on neuroimages. In each patient, the total number of days with TNEs was recorded and defined as the duration of the postoperative TNEs.

2.4. Statistical analysis

Data were expressed as mean ± standard deviation, and median ± interquartile range, as appropriate. The Fisher's exact test and Wilcoxon rank sum test were used to compare the SCLI and CHB scores between patients with and without TNEs, respectively. Sensitivity and specificity were estimated for different sets of cut-off values. To minimize the potential bias to select a single cut-off value for positivity, a receiver operating characteristic curve was constructed. Analysis was performed using commercially available software (JMP 9 [SAS Institute Inc., Cary, North Carolina, USA]). P values < 0.05 were considered statistically significant.

3. Results

There was no case that had the SCLI or CHB signs on preoperative FLAIR images. Double direct bypass was performed in 3 (16.7%) hemispheres of 3 (18.8%) cases. The patency of direct anastomosis was confirmed with postoperative MR angiography in all cases.

3.1. TNEs after revascularization surgery

Postoperative TNEs occurred derived from 8 (44.4%) hemispheres in surgically treated cases. Major clinical symptoms were numbness of the extremities (11.1%), aphasia (33.3%), facial palsy (11.1%), dysarthria (11.1%), hemiparesis (22.2%), and seizure (11.1%). The median time of surgery to TNE occurrence was 3 days, and the average duration was 5.0 ± 2.5 days. TNEs occurred more frequently after left-sided surgery than right-sided surgery (6 vs. 2, respectively). Local hyperperfusion was observed in one case, which presented with a TNE. There was no significant difference in the occurrence of TNE between single and double bypass procedures (33.3% vs. 46.7%, respectively).

3.2. SCLI and CHB signs

Postoperative SCLI and CHB signs on FLAIR images were detected in 9 (50.0%) and 12 (66.7%) hemispheres in surgically treated cases, respectively. The SCLI scores were 0 in 9 cases (50.0%), 1 in 1 cases (5.6%), 2 in 3 cases (16.7%), 3 in 3 cases (16.7%), and 4 in 2 cases (11.1%). The CHB scores were 0 in 6 cases (33.3%), 1 in 1 case (5.6%), 2 in 3 cases (16.7%), 3 in 4 cases (22.2%), and 4 in 4 cases (22.2%). Neither acute cortical infarction (diffusion weighted MR images-