Novel Indirect Revascularization Technique with Preservation of Temporal Muscle Function for Moyamoya Disease Encephalo-Duro-Fascio-Arterio-Pericranial-Synangiosis: A Case Series and Technical Note

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■ BACKGROUND: **Direct and/or indirect bypass surgery is the established approach for preventing stroke in patients with moyamoya disease. However, conventional indirect revascularization, including encephalo-myo-synangiosis, has some disadvantages associated with the mass effect of the temporal muscle under the bone flap and postsurgical depression in the temporal region. We devised a novel indirect revascularization method, using only the temporal fascia, to address the aforementioned disadvantages.**

■ METHODS: **A skin incision was performed along the superficial temporal artery. The temporal fascia was cut such that the base of the fascia flap was on the posterior side. The fascia and temporal muscles were dissected separately. After turning over the fascia, the muscle was cut such that the base of the muscle flap was on the anterior side. Craniotomy, direct bypass, and encephalo-duro-synangiosis were performed conventionally. Only the temporal fascia was used for indirect revascularization and duraplasty. The muscle was replaced in the anatomically correct position after replacing the bone flap.**

■ RESULTS: **We performed the aforementioned surgery on 18 (13 women and 5 men) consecutive patients (21 cerebral hemispheres) enrolled between 2012 and 2016. The average age was 28.7 years. The mean follow-up period was 31.6 months. In 17 patients (94%), the symptoms and cerebral blood flow improved. Digital subtraction angiography showed satisfactory angiogenesis from the temporal fascia. Depression in the temporal region and atrophy of the temporal muscle were negligible.**

■ CONCLUSIONS: **This surgical technique provides good clinical and cosmetic outcomes. It may also be one of the good surgical treatments available for symptomatic moyamoya disease.**

Moyamoya disease is characterized by idiopathic progressive arterial stenosis or occlusion of the circle of Willis and development of an abnormal fragile vascular network, frequently causing stroke in children or young adults. The symptoms are transient ischemic attacks, cerebral infarction, and hemorrhage.[1-4] To prevent future instances of stroke, direct and/or indirect bypass surgical procedures have been established for patients with moyamoya disease.[5-11] Encephalo-myo-synangiosis (EMS), using the temporal muscle, is one of the major indirect procedures. EMS was first described by Karasawa et al.,[12] and many studies have reported the effectiveness of EMS as an indirect procedure for treating moyamoya disease.[6,13] However, the conventional methods of indirect revascularization, including EMS and similar techniques, such as encephalo-duro-arterio-myo-synangiosis, have some serious disadvantages. First, these techniques lead to a mass effect of the temporal muscle under the bone flap, and some studies have reported the calcification or acute swelling of the muscle flap under the bone flap, causing focal neurologic deficits and convulsive seizure.[14-16] Second, insertion of the temporal muscle under the bone flap and wide temporal bone window to prevent the compression of the vascularized temporal muscle flap causes postsurgical depression in the temporal region.

Key words
- Combined revascularization
- Cosmetic
- Moyamoya disease
- Surgical treatment
- Technical note
- Temporal muscle

Abbreviations and Acronyms
CBF: Cerebral blood flow
DSA: Digital subtraction angiography
EMS: Encephalo-myo-synangiosis
MCA: Middle cerebral artery
MRI: Magnetic resonance imaging
mRS: Modified Rankin Scale
SPECT: Single photon emission computed tomography
STA: Superficial temporal artery

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These are serious issues because there are many young children and female patients with moyamoya disease. To solve these problems, we devised a new surgical method of indirect revascularization that uses only the temporal fascia, and not the temporal muscle.

MATERIALS AND METHODS

Patient Population

This surgical method was used to treat 18 consecutive patients (13 women and 5 men) with moyamoya disease (21 cerebral hemispheres) who visited our institute between April 2012 and March 2016. The average age was 28.7 ± 16.7 years (range, 3–56 years). Eleven patients (61%) were treated only on the right side, and 4 patients (22%) were treated only on the left side. Both hemispheres were treated in 3 patients (17%). In one of these cases, both hemispheres were treated in a single step. Clinical presentations included transient ischemic attacks in 11 patients (61%), ischemic stroke in 3 patients (17%), hemorrhagic stroke in 2 patients (11%), and periodic headaches in 2 patients (11%). Fifteen patients (83%) had a preoperative modified Rankin Scale (mRS) score of 0–1, 2 patients (11%) had a score of 2, and 1 patient (6%) had a score of 4. Patient characteristics and clinical presentation are shown in **Table 1**. All patients were symptomatic and showed reduced cerebral blood flow (CBF) in the affected side, detected using single photon emission computed tomography (SPECT). All cases were diagnosed as moyamoya disease using well-established and generally accepted diagnostic criteria.[17,18] We obtained informed consent in all cases, and the institutional review board approved the study protocol (number 17183). In patients younger than 18 years of age, informed consent was obtained from the parent or guardian.

Table 1. Demographic Data and Clinical Presentation (N = 18)

Characteristic	Value
Mean age ± SD (range) (years)	28.7 ± 16.7 (range, 3–56)
Sex	
Female	13 (72)
Male	5 (28)
Total number of hemispheres treated	21
Unilateral treatment	15 (83)
Treatment for right side only	11 (61)
Treatment for left side only	4 (22)
Bilateral treatment	3 (17)
Clinical presentation	
Periodic headache	2 (11)
TIA	11 (61)
Ischemic stroke	3 (17)
Hemorrhagic stroke	2 (11)
Preoperative mRS score	
0–1	15 (83)
2	2 (11)
4	1 (6)

Values are numbers (%) unless indicated otherwise.
TIA, transient ischemic attack; mRS, modified Rankin Scale.

Surgical Outcome

After performing the surgery, we followed-up the patients and recorded their clinical presentations. All adverse events experienced by the patients were recorded without exception. Magnetic resonance imaging (MRI) and magnetic resonance angiography were performed within 3 days of surgery to evaluate the surgical complications and confirm the bypass patency; SPECT was performed within 3 days of surgery and a period of 6 months to 1 year after surgery to assess the improvement in CBF. The mRS scores were evaluated 1 year after surgery. To evaluate angiogenesis, we performed digital subtraction angiography (DSA) within 6–12 months of surgery. To evaluate cosmetic outcome for the patients for whom the surgery was performed on only one cerebral hemisphere and more than 2 years had passed since the surgery, we compared the thickness of the temporal muscle on the bone flap of the operative side and that of the nonoperative side at the level of the foramen of Monro in MRI axial slices.

Surgical Technique

The patient was placed in a supine position with the head rotated between 30° and 60° to the opposite side of the surgical site in a Mayfield headrest. The temporal region was kept horizontal using a shoulder pillow. A skin incision was made along the parietal branch of the superficial temporal artery (STA) and extended forward in a gently curving arc. After dissecting the parietal branch of the STA, an additional skin incision was made and extended backward from a point where it intersected the temporal line (**Figure 1A**). The frontal branch of the STA was dissected from the inner side of the skin flap in cases where it was necessary. This skin incision provided us with a wide access to the temporal fascia and frontal periosteum (**Figure 1B**), and we could use frontotemporal craniotomy to perform indirect revascularization for a wide region. The temporal fascia was cut such that the base of the vascularized fascia flap was on the posterior side, and the temporal fascia and the temporal muscle were dissected separately. After turning over the temporal fascia (**Figure 1C**), the temporal muscle was cut such that the base of the vascularized fascia flap was on the anterior side. The temporal fascia on the temporal line was sutured with the reconstructed temporal muscle after replacing the bone flap. The frontal periosteum was also cut to create a vascularized flap with its base directed toward the anterior side (**Figure 1D**). After turning over these vascularized flaps, a wide craniotomy was carefully performed so as not to damage the middle meningeal artery. The dura mater was cut open to form dural flaps, while retaining the main branch of the middle meningeal artery. A suitable cortical recipient artery (more