

Invited Article: Surgical Management of Moyamoya Disease

Moyamoya Hastalığının Cerrahi Tedavisi

ABSTRACT

OBJECTIVE: Various types of revascularization surgery have been performed for moyamoya disease, and there have been controversies about the surgical procedures. In this report, we introduce our surgical strategy of moyamoya disease and discuss the various surgical procedure options.

METHODS: The subjects consisted of 13 consecutive patients (8 children, 5 adults) with moyamoya disease who were surgically treated between 2004 and 2007. The onset symptoms were ischemia (11 patients), hemorrhage (1 patient), or headache (1 patient). We performed modified standard encephalo-duro-arterial-synangiosis on 16 sides in the 8 children and superficial temporal artery-to-middle cerebral artery anastomosis with encephalo-duro-myosynangiosis combining direct and indirect bypasses on 5 sides in the 5 adult patients.

RESULTS: Perioperative complications were noted in 12 (9 patients) of 21 operations (13 patients). Most complications were transient and no attributive lesions were detected on CT or MRI. The clinical outcome was excellent or good and revascularization was seen in cerebral blood flow studies. Effective neovascularization through the grafts was observed in follow-up angiography.

CONCLUSIONS: The surgical management of moyamoya disease has various aspects depending on the individual subject and very specific surgical management might be required.

KEY WORDS: Bypass surgery, Cerebrovascular disease, Circle of Willis, Hemorrhage, Ischemia, Moyamoya disease, Stroke, Surgery.

ÖZ

AMAÇ: Moyamoya hastalığı değişik tiplerde yapılabilen revaskülarizasyon cerrahisi ile tedavi edilmektedir, ancak cerrahi yöntemler ile ilgili olarak kesin bir fikir birliği oluşmamıştır. Bu çalışmada bizim uyguladığımız cerrahi yöntem ile halen uygulanmakta olan cerrahi yöntemler tartışılmaktadır.

YÖNTEM: Serimiz Moyamoya hastalığı nedeni ile 2004- 2007 yılları arasında ardışık olarak opere edilen 13 hastadan oluşmaktadır (8 çocuk, 5 yetişkin). Başlangıç belirtisi: 13 hastanın 11'inde iskemi, 1 hastada kanama ve 1 hastada ise baş ağrısı olarak tesbit edildi. Serimizde teknik olarak modifiye standart encephalo-duro-arterial synangiosis uygulaması yapılmıştır. 8 çocukta iki taraflı olarak superfisiyel temporal arter ile orta serebral arter arasında anastomozu yapılmıştır, aynı işlem 5 erişkinde tek taraflı olarak yapılmıştır.

SONUÇLAR: 13 hastaya toplam 21 operasyon yapıldı 9 hastada 12 komplikasyon gelişti, komplikasyonların çoğu geçici ve bilgisayarlı tomografi ve manyetik görüntüleme operasyona ikincil olarak gelişen herhangi bir lezyon saptanmamıştır. Klinik sonuçlar mükemmel ya da mükemmele yakın olarak bulunmuştur, aynı zamanda serebral kan akımı çalışmalarında revaskülarizasyonun geliştiği tesbit edilmiştir. Etkili bir neovaskülarizasyonun geliştiği takip angiografiler ile gösterilmiştir.

SONUÇ: Moyamoya hastalığının cerrahi tedavisi hastaların özellikleri gözönüne alınarak yapılması gereken ve nadiren çok spesifik cerrahi tekniklere ihtiyaç duyulan bir durumdur.

ANAHTAR SÖZCÜKLER: Bypass cerrahisi, Cerebrovasküler hastalık, Cerrahi, inme, İskemi, Kanama, Moyamoya hastalığı, Willis halkası

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INTRODUCTION

Moyamoya disease is a progressive occlusive cerebrovascular disease characterized by bilateral stenosis of the internal carotid arteries (ICAs) and the development of compensatory collateral vessels (31, 32, 33). Various surgical procedures have been applied to this condition to prevent ischemic deficits, including direct bypass procedures such as superficial temporal artery-to-middle cerebral artery (STA-MCA) anastomosis (13, 19); indirect bypass procedures such as encephalo-duro-arterio-synangiosis (EDAS) (25), encephalo-duro-arterio-myosynangiosis (EDAMS) (18), encephalo-myosynangiosis (EMS) (12), omental transplantation onto the cortical surface (15), multiple burr holes (2, 16), and a combination of direct and indirect bypasses (5, 9, 12, 14, 18) have also been used. Though the efficacy of these revascularization surgeries for moyamoya disease has been well recognized, the optimal surgical procedure remains unknown.

In this report, we detail the use of modified encephalo-duro-arterial-synangiosis (EDAS) on 16 sides in 8 pediatric patients and STA-MCA anastomosis with encephalo-duro-myosynangiosis (EDMS) modified to combine direct and indirect bypasses on 5 sides in 5 adult patients. We introduce the operative procedures and discuss the surgical management of moyamoya disease based on our experience and the published literature.

PATIENTS and METHODS

Patient Profile

The study population consisted of 13 consecutive patients (6 males, 7 females) with moyamoya disease who were surgically treated between 2004 and 2007. The age of the patients at the time of initial operation ranged from 5 to 48 years (8 children, 5 adults). Onset symptoms were transient ischemic attack (TIA) (8 patients), infarction (3 patients), hemorrhage (1 patient), or headache (1 patient). Six patients with TIA or headache presented with no symptoms on initial admission, while one patient with TIA showed an asymptomatic infarction area on computed tomography (CT) and magnetic resonance imaging (MRI). The angiographic stages, according to Suzuki and Takaku (32), were Stage 3 or 4. The detailed characteristics of the 13 patients are summarized in Table 1.

Surgery

We performed encephalo-duro-arterial-synangiosis (EDAS) on 16 sides in 8 pediatric patients and STA-MCA anastomosis with encephalo-duro-myosynangiosis (EDMS) on 5 sides in 5 adult patients. The EDAS was performed based on the method of Matsushima et al. (25) with some modifications, and STA-MCA anastomosis with EDMS was performed with modifications to combine direct and indirect bypasses (5, 12, 14, 18). In the pediatric patients, the interval between the operations on each side was between 2 and 10 months (mean interval: 5 months).

Surgical Procedure

Modified EDAS (Figure 1): A skin incision was made along a parietal branch of the STA and extended upward to the anterior by approximately 4 cm, using caution to not cut an anterior branch of the STA. The galeal flap with a parietal branch of STA was dissected as a graft. The temporal muscle was cut crosswise, and a fronto-temporal craniotomy (7-8 cm height and 7-8 cm width) was performed. The dura mater was cut without severing a main branch of middle meningeal artery (MMA), and the dura mater was then turned over and attached to the brain surface. The galeal flap with a parietal branch of the STA was attached to the surface of cortex and sutured to the dura mater.

STA-MCA anastomosis with encephalo-duro-myosynangiosis (EDMS) (Figure 2): A skin incision was made along a parietal branch of the STA and extended upward to the anterior by approximately 6 cm, using caution to not cut an anterior branch of the STA. A parietal branch of the STA was dissected as a donor vessel for direct bypass. The temporal muscle was cut as wide as possible along the skin incision and linear temporalis, and a fronto-temporal craniotomy (7-8 cm height and 9-10 cm width) was performed. The dura mater was cut without severing a main branch of middle meningeal artery (MMA), and the dura mater was turned over and attached to the brain surface. The STA-MCA bypass was then performed; a dissected temporal muscle used as a graft was attached to the surface of cortex and sutured to the dura mater.

Perioperative Complications

Perioperative complications were noted in 12 of 21 operations (9 of 13 patients). In 11 operations (8 patients), the symptoms were transient and no

Table 1: The characteristics of the cases

Case No.	Age /sex at initial operation years	Type of onset (side)	Presenting symptoms on admission	CT /MRI findings on admission	Angiographical stage	Operative procedure (side)	Perioperative complications (side)	Follow-up period	Clinical outcome
1	48/M	Infarction (R)	Dysarthria	Infarction (R. frontal)	R: 4, L: 3	STA-MCA bypass + EDMS (R)	None	3 years	Excellent
2	5/M	TIA (R) Infarction (R), TIA (L)	None L. hand weakness	Normal Infarction (B. frontal)	R: 3, L: 3	EDAS (R) EDAS (L)	Infarction (R) None	3 years	Good
3	13/F	TIA (R)	L. arm involuntary movement	Normal	R: 3, L: 3	EDAS (R)	TIA (L)		
4	8/F	None TIA (L) TIA (R)	Normal None None	Normal Normal	R: 3, L: 3	EDAS (L) EDAS (L) EDAS (R)	None None TIA (R)	3 years	Excellent
5	9/F	TIA (R and L)	None	Infarction (R. parieto-occipital) Normal	R: 5, L: 4	EDAS (R) EDAS (L)	None TIA (L)	3 years	Excellent
6	17/F	Headache Headache	None None	Normal Normal	R: 4, L: 4	EDAS (L) EDAS (R)	TIA (L) TIA (R)	3 years	Excellent
7	16/F	TIA (L) TIA (R)	None None	Normal Normal	R: 5, L: 5	EDAS (L) EDAS (R)	TIA (L) TIA (R)	2 years	Excellent
8	5/M	Infarction (R) TIA (L)	L. arm weakness Infarction (R. frontal)	Infarction (R. frontal)	R: 3, L: 3	EDAS (R) EDAS (L)	TIA (R-L) TIA (L)	2 years	Good
9	14/M	TIA (L) None	R. facial and arm weakness Normal	Normal	R: 4, L: 4	EDAS (L) EDAS (R)	TIA (L) None	1 year	Excellent
10	22/F	TIA (R)	L. arm involuntary movement	Normal	R: 4, L: 1	STA-MCA bypass + EDMS (R)	None	1 year	Excellent
11	27/F	Hemorrhage Infarction (L)	Consciousness disturbance Total aphasia, R. hemiparesis	Intraventricular hemorrhage Infarction (L. fronto-parietal)	R: 1, L: 3	STA-MCA bypass + EDMS (L)	None	1 year	Good
12	34/M	Infarction (L)	Motor aphasia	Infarction (L. frontal)	R: 3, L: 4	STA-MCA bypass + EDMS (L)	None	4 months	Excellent
13	23/M	TIA (L)	None	Normal	R: 1, L: 4	STA-MCA bypass + EDMS (L) TIA (L)	None	4 months	Excellent

TIA: Transient ischemic attack, R: Right, L: Left.

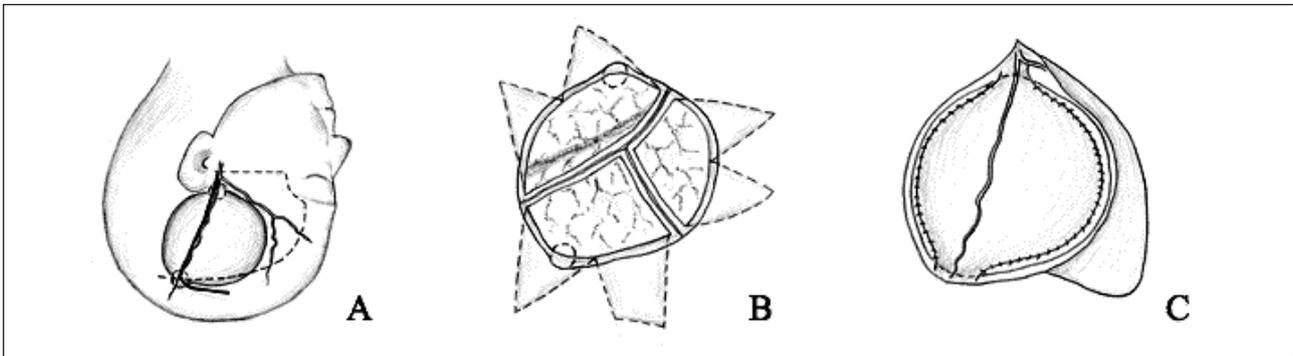


Figure 1: Schematic drawing of the operative procedure of encephalo-duro-arterio-synangiosis with some modification: A skin incision is made along a parietal branch of the superior temporal artery (STA) and extended upward to the anterior approximately 4 cm, using caution to not cut an anterior branch of the STA (A). The galeal flap, with a parietal branch of the STA, is dissected as a graft. The temporal muscle is cut crosswise, and a fronto-temporal craniotomy (7-8 cm height and 7-8 cm width) is performed (A). The dura mater is cut without severing a main branch of the middle meningeal artery (MMA); the dura mater is then turned over and attached to the surface of cortex (B). The galeal flap, with a parietal branch of the STA, is attached to the surface of cortex and sutured to the dura mater (C).

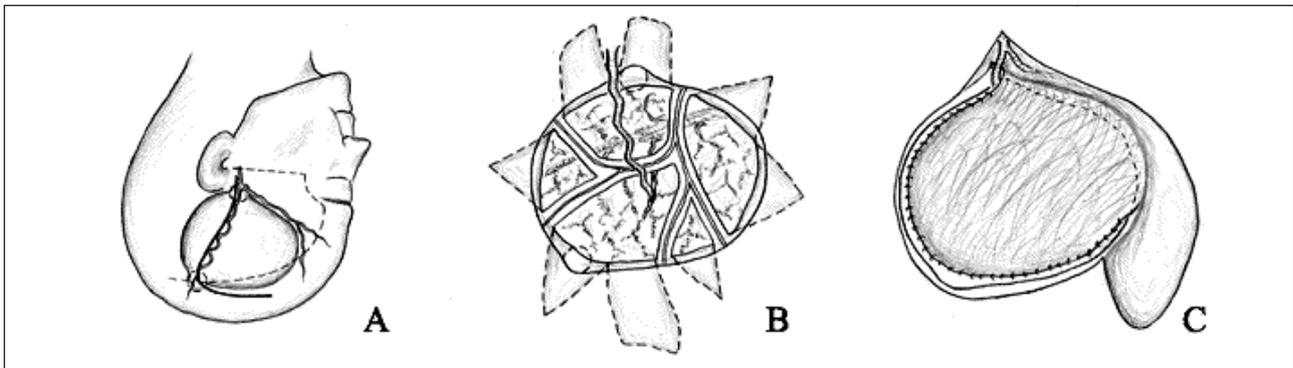


Figure 2: Schematic drawing of the operative procedure of superior temporal artery (STA)-to-middle cerebral artery (MCA) anastomosis with encephalo-duro-myo-synangiosis (EDMS): A skin incision is made along a parietal branch of the STA and extended upward to the anterior approximately 6 cm, using caution to not cut an anterior branch of the STA (A). A parietal branch of the STA is dissected as a donor for the direct bypass. The temporal muscle is cut as wide as possible along the skin incision and linear temporalis, and a fronto-temporal craniotomy (7-8 cm height and 9-10 cm width) is performed (A). The dura mater is cut without severing a main branch of the middle meningeal artery (MMA); the dura mater is then turned over and attached to the surface of the cortex (B). The STA-MCA bypass is performed, and a dissected temporal muscle used as a graft is attached to the surface of the cortex and sutured to the dura mater (B and C).

attributive lesion was detected on CT or MRI. However, a 5-year-old boy (Case 2) developed slight left hand weakness due to post-operative crescendo ischemia of the operative side. To date, these 13 patients have been followed up by neurological and radiological examinations, including angiography and cerebral blood flow (CBF) measured by single photon emission CT using 123I-iodoamphetamine (IMP) and 99mTc- d,l-hexamethyl-propyleneamine oxime (HM-PAO).

RESULTS

Clinical Status

Of the 13 patients studied, 10 patients had no

post-operative neurological deficit. Of the three other patients, one (Case 2) showed slight left hand weakness, another (Case 8) showed slight left arm weakness, and the third patient (Case 11) had slight right hemiparesis. In these three cases, the patients had these neurological deficits on admission (Cases 8 and 11) or perioperative complications induced the deficit (Case 2). However, these three patients continue to do well in their daily lives.

Follow-Up CT/MRI Findings

There were no remarkable abnormal findings in CT or MRI for seven patients. Of the six other patients, there was cerebral infarction (Cases 1, 2, 5,

8, and 12) or intraventricular hemorrhage (Case 11) observed from CT or MRI images at admission. However, there were no new abnormal lesions reported after discharge.

Follow-Up CBF Findings

Cerebral blood flow studies were performed for all patients preoperatively. In 12 of 13 patients (except Case 8), CBF studies were also performed from 7 days to 22 months postoperatively. In all patients who underwent CBF studies, blood flow reserve in the frontal and parietal lobe of operative side were improved (excluding the infarction area).

Follow-Up Angiographic Findings

Angio graphic studies were performed for all patients preoperatively. In 11 of 13 patients (except Cases 2 and 8), angio graphic studies were also performed from 9 days to 28 months postoperatively. In the six pediatric patients who underwent only indirect bypass surgery, angio graphic studies of both sides were performed after the operation, and well-developed neovascularization of the grafted tissue was observed. The superficial temporal artery, deep temporal artery, and middle meningeal artery were the usual donors for neovascularization. In the five adult patients who underwent direct and indirect bypass surgery, the interval from operation to the follow-up angiographic studies ranged from 9 days to 6 months; collateral circulation was mainly supplied through the direct bypass procedure.

DISCUSSION

Various types of revascularization surgery have been used to treat patients with moyamoya disease (2, 5, 9, 12-16, 18, 19, 25), with some controversy about the most appropriate surgical procedures. The greater advantage of either direct bypass surgery or indirect bypass surgery remains unresolved. In some previous studies, the advantage of direct bypass surgery has reportedly depended on the blood supply available immediately after surgery, and perioperative complications are more likely in cases treated by indirect than direct bypass surgery (14, 24). However, in other studies, indirect bypass surgery might be more advantageous due to its simple and less-invasive nature (18, 25, 30). Each surgical procedure has advantages and disadvantages, and one of the disadvantages of EDAS or EDAMS is its restriction to only the area of the MCA.

Knowledge of the natural history of moyamoya disease is necessary in determining appropriate surgical management. Analysis of the natural development of the disease is difficult because the majority of cases are treated surgically. However, the natural history of pediatric moyamoya disease has been investigated; in certain reports, more than half of the patients not treated surgically had neurological deficits due to recurrent ischemic attacks (22, 30). In addition, a recent report from Japan indicated that asymptomatic moyamoya disease is not a silent disorder and may potentially cause ischemic or hemorrhagic stroke (21). In previously reported studies, the efficacy of revascularization surgery has been recognized in preventing ischemic insults, especially in pediatric patients (1, 3, 6, 8, 10, 14, 19, 22, 27, 29, 30). In adult patients with moyamoya disease, the effect of indirect bypass procedures remains controversial compared to that of direct bypass surgery (11, 19, 26, 28). On the other hand, efficacy in preventing future hemorrhage is still controversial (4, 6, 17, 20). The reduction of abnormal dilated collateral vessels, including aneurysm formation, has been considered an important factor in preventing future hemorrhage (20, 27).

Our previous study indicated that the majority of moyamoya disease cases progress quickly during early childhood and become stable later in childhood and adolescence (based on temporal profiles of angio graphic findings) (7). In addition, our previously reported data indicated that certain pediatric cases where the disease appeared after the patients reached 5 years of age presented with stable temporal changes in angio graphic stage; this finding might indicate that in only a few adult cases does the disease initiate in adulthood (7). Thus, we must perform revascularization surgery in disease's acute stage, before the appearance of permanent neurological deficits.

Accurate analysis of the hypoperfusion area and comparison to clinical symptoms is significant process in determining the revascularization area and surgical procedure. Our alternative surgical strategy depends on our experience and previously reported cases. In pediatric patients, we performed modified standard EDAS as the first choice, because this method was simple, safe, less invasive, less time-consuming, and comparatively effective (1, 3, 6, 10, 29). Furthermore, a less invasive and less time-

consuming operative procedure was required to prevent perioperative complications. Though we performed the operation in separate stages on each side, it would be possible to operate on both sides simultaneously. In adult patients, we performed STA-MCA anastomosis with modified EDMS to combine direct and indirect bypasses, because good revascularization was not expected in indirect surgery (11, 19, 26, 28). In all cases, when the hypoperfusion area and clinical symptoms appeared, we perform additional procedures to the affected area.

In this updated case series, postoperative clinical outcome has been good even with limited short-term follow-up. We previously reported that good revascularization seen in most patients over long-term follow-up (3, 10). In a few cases, revascularization was poor, particularly in the anterior cerebral artery (ACA) territory, as reported by other authors (9, 23). In our initial surgical management, the anterior branch of STA were preserved for the additional option of a direct bypass to ACA territory.

In conclusion, the surgical procedure for moyamoya disease must be selected depending on clinical symptoms, CBF reserve, and individual surgeon preference. In the future, tailor-made surgical management adjusted to individual variations might be required.

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