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Multiple EDAS (encephalo-duro-arterio-synangiosis)

Additional EDAS using the frontal branch of the superficial temporal artery (STA) and the occipital artery for pediatric moyamoya patients in whom EDAS using the parietal branch of STA was insufficient

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Abstract Although parietal EDAS or STA-MCA anastomosis are effective in pediatric moyamoya disease, they do not adequately prevent ischemia in the frontal and occipital lobes. Some additional methods that can prevent ischemia in the frontal and occipital lobes are sometimes needed. We investigated whether EDAS using a frontal branch of the superficial temporal artery (frontal EDAS) or EDAS using the occipital artery (occipital EDAS) is preferable. Frontal or occipital EDAS was performed at 15 sites in seven patients with pediatric moyamoya disease. The outcome was estimated by angiography 3 months later, CT findings 3 months later, neurological findings during the follow up period and perioperative complications. The mean follow up period was 14±6 months after frontal or occipital EDAS. As results, good revascularization from frontal or occipital EDAS was shown in ten of fourteen surgical sites (71%) in angiography. None of the patients showed deterioration of symptoms after frontal or occipital EDAS during the follow up period. None of the patients developed surgical complications. In conclusion, multiple EDAS using the frontal branch of STA and the occipital artery is an effective and safe method for preventing ischemia in the frontal and occipital lobe in pediatric moyamoya disease.

Key words Moyamoya disease · Encephalo-duro-arterio-synangiosis (EDAS) · Frontal lobe · Occipital lobe

Introduction

Reconstructive vascular surgery is recommended for pediatric moyamoya disease [2], and EDAS (encephalo-duro-arterio-synangiosis) using the parietal branch of the superficial temporal artery (STA) [7, 18], and STA–MCA anastomosis with EMS (encephalo-myo-angiosis) [8] are common methods. Although these methods are effective, they do not provide adequate prevention of ischemia in the frontal and occipital lobes [4, 23].

Consequently, additional methods that can prevent ischemia in the frontal and occipital lobes are needed. In this study, we investigated whether EDAS using a frontal branch of the STA (frontal EDAS) or EDAS using the occipital artery (occipital EDAS) is preferable. We performed frontal or occipital EDAS in seven patients and discuss the usefulness of this technique.

Patients and methods

Frontal or occipital EDAS was performed at 15 sites in seven patients with pediatric moyamoya disease. The diagnosis was based on the guidelines of the Research Committee on Spontaneous Occlusion of the Circle of Willis (Moyamoya Disease) of the Ministry of Health and Welfare, Japan [12]. The patients' ages ranged from 1 to 12 years. EDAS using the parietal branch of STA had previously been performed in all patients. Clinical symptoms, follow-up angiography, follow-up CBF (cerebral blood flow) study (Tohoshiba

GCA-901A, Toshiba, Tokyo), and follow-up CT were evaluated in each case. Indications for frontal or occipital EDAS were as follows: additional symptoms, low CBF in the frontal or occipital area, frontal or occipital lobe atrophy on CT. Surgery was performed under general anesthesia induced and maintained by skilled anesthesiologists. The frontal branch of STA or the occipital artery was dissected around 5 cm length from the hairline or nuchal line. The strip was sutured to the dura mater [18] with as little damage to the collateral circulation as possible. The outcome was estimated with reference to angiography 3 months later, CT findings recorded 3 months later, neurological findings recorded during the follow-up period, and perioperative complications. Angiographic findings were divided into three categories: good (revascularization was seen to extend beyond the EDAS area), fair (revascularization was restricted to the EDAS area), and none (no revascularization). A DQ/IQ (developmental/intelligence quotient) under 75 [20] was considered to constitute mental retardation. The mean follow-up period was 14±6 months after frontal or occipital EDAS.

Illustrative case reports

Case 1

A 12-year-old boy developed hemiparesis in 1987. Angiography showed moyamoya vessels. Bilateral parietal EDAS was performed. The postoperative course was uneventful. Because CBF study showed low perfusion in the bilateral frontal lobes and the boy developed borderline mental retardation, bilateral frontal EDAS was performed in 1995. Angiography of the bilateral external carotid artery showed good revascularization from both parietal EDAS and frontal EDAS (Fig. 1A). The postoperative course was uneventful and the patient is currently attending a junior high school.

Case 5

A 2-year-old girl developed hemiparesis in 1993. Angiography showed moyamoya vessels. Bilateral parietal EDAS was performed. After the initial surgery, left parietal EDAS showed poor revascularization (Fig. 2A) and periodic unstable gait persisted. For these reasons, left frontal EDAS was added. Frontal EDAS and parietal EDAS developed after the second procedure (Fig. 2B). The patient is currently attending a kindergarten.

Case 6

A 1-year-old infant girl developed hemiparesis in 1994. Angiography showed moyamoya vessels. Bilateral parietal EDAS was performed. After the initial surgery, left subdural hemorrhage developed. Unstable gait attack continued, and CBF showed bilateral frontal and left occipital low perfusion; therefore, bilateral frontal and occipital EDAS was performed. Although frontal EDAS led to only fair revascularization, parietal and occipital EDAS showed good revascularization (Fig. 3A). The patient is currently attending a kindergarten.

Results

The results are summarized in Table 1.

Angiography revealed good revascularization from frontal or occipital EDAS in 10 of 14 surgical sites (71%) and fair revascularization in 4 of 14 sites (28%).

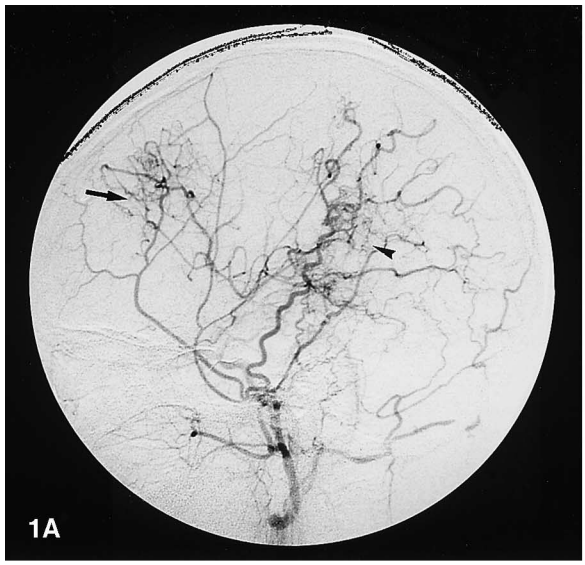
CT showed similar findings before surgery in all patients. None of the patients had additional low-density areas after the final surgery (frontal and/or occipital EDAS).

Neurological monitoring did not reveal worsening of symptoms or ischemic attacks after the final surgery (frontal and/or occipital EDAS) in any of the patients during the follow-up period. Two patients (cases 1, 2), in whom frontal EDAS was added 5 and 6 years after parietal EDAS, had borderline mental retardation before frontal EDAS. In case 3, the patient complained of periodic unstable gait after parietal EDAS, and frontal and occipital EDAS were added for this reason. The symptom disappeared postoperatively. In case 4, frontal EDAS was added after parietal EDAS because of persistent urinary incontinence. The symptom disappeared after frontal EDAS. In two patients (cases 5, 6), we added frontal EDAS because of unstable gait attacks and later added occipital EDAS after frontal EDAS because periodic unstable gait persisted. The symptom disappeared after occipital EDAS. The remaining patient (case 7) underwent parietal and frontal or occipital EDAS within a short time and did not show any neurological deterioration or mental retardation.

None of the patients developed surgical complications.

Discussion

EDAS using the parietal branch of the STA [7, 18] and STA–MCA anastomosis with EMS [8, 9] are effective methods of reconstructive vascular surgery for pediatric moyamoya disease [2], but do not always adequately prevent ischemia in the frontal and occipital lobes, as demonstrated by CBF [4, 23]. Consequently, other methods are often necessary to increase CBF in the frontal or occipital lobes. Omental transplantation [10], ribbon EDAS [11], EMAS (encephalo-myo-arterio-synangiosis [5, 16, 17, 21], and direct anastomosis [3, 6, 22] have been reported as methods of revascularization in the frontal or occipital lobe. The technical difficulties, which include sacrifice of distal collateral circulation from the frontal branch of STA and occipital artery, may be disadvantages of these methods. Generally, STA–MCA anastomosis causes ischemic complications in around 4% of cases [1], so that these sophisticated but complex methods can have some perioperative complications [24]. Furthermore, in pediatric moyamoya disease, prophylactic treatment is often required [14]. In prophylactic surgery, perioperative complications should be minimized. In EDAS distal collateral circulation is rarely sacrificed, because the skin incision is linear and the distal frontal branch of the STA and the occipital artery are preserved [18]. Because of the simplicity of the procedure, frontal EDAS and occipital EDAS are recommended. In our series no perioperative complications de-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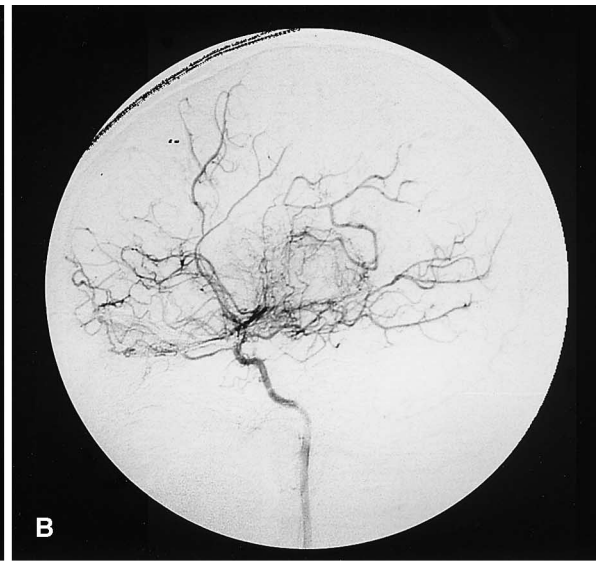
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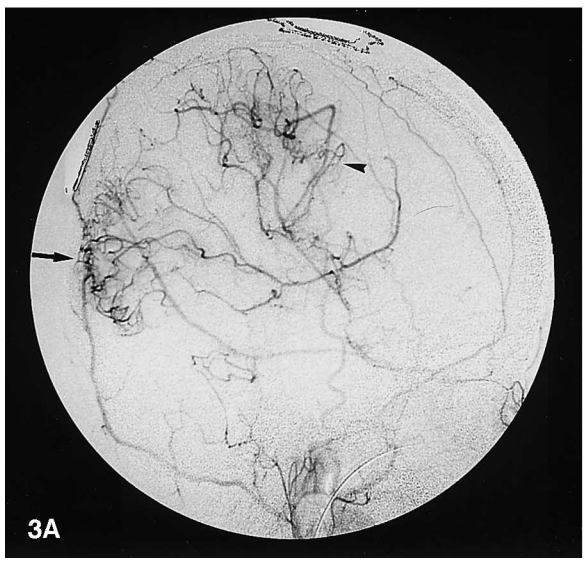
2A


B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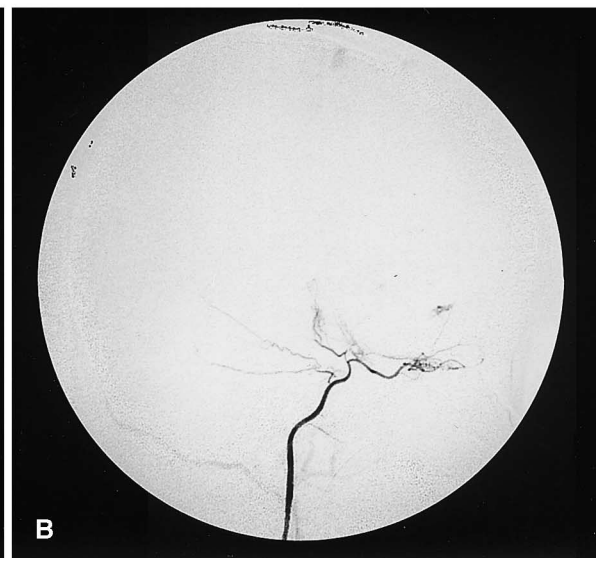
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Table 1 Treatment by encephalo-duro-arterio-synangiosis (EDAS) and outcome in seven children with moyamoya disease (*R* right, *L* left, *B/Bil* bilateral, *P* EDAS using superficial temporal artery [STA] parietal branch, *F* EDAS using STA frontal branch, *O* EDAS using occipital artery, *good* revascularization beyond operative area, *fair* revascularization limited to operative area, *none* no revascularization)

Case no.	Onset		Operation			Outcome	
	Age (year)	Symptom	Methods and date	Perioperative complications	Postoperative angiography	Age, CT	Symptoms
1	4 (1987)	L. hemiparesis	R. P. (1987/12) L. P. (1988/8) B. F. (1996/1)	None None None	Good Good Good	12, no findings	None
2	2 (1989)	Bil. hemiparesis	R. P. (1990/4) L. P. (1990/8) B. F. (1996/3)	None None None	Good Good Good	8, no findings	None
3	1 (1993)	R. monoparesis	L. P. (1993/7) R. P. (1993/8) R. F. O. (1995/10)	None None None	Good None Fair	4, R. parietal infarction	None
4	8 (1993)	Drop attack	L. P. (1994/3) R. F. (1994/9)	None None	Good Good	10, no findings	None
5	2 (1993)	L. hemiparesis	R. P. (1994/5) L. P. (1994/8) R. F. (1995/7) L. O. (1995/12)	None None None None	Fair Good Good –	4, L. occipital infarction	R. hemiparesis (mild)
6	1 (1994)	R. hemiparesis	L. P. (1994/10) R. P. (1994/11) R. L. F. (1995/7) R. L. O. (1996/4)	None None None None	Fair Good Fair Good	3, L. hemispheric infarction	None
7	4 (1996)	L. hemiparesis	R. P. O. (1996/3) L. P. F. (1996/3)	None None	Good Good	4, R. parietal infarction	None

◄ **Fig. 1A, B** Case 1. **A** Angiography of left external carotid artery. *Arrowhead* shows previous parietal encephalo-duro-arterio-synangiosis (EDAS). *Arrow* shows frontal EDAS. **B** Angiography of left internal carotid artery

Fig. 2A, B Case 5. **A** Angiography of right external carotid artery. *Arrowhead* shows previous parietal EDAS. Poor revascularization is revealed. **B** Angiography of right external carotid artery after frontal EDAS was added. *Arrow* shows frontal EDAS covered frontal lobe

Fig. 3A, B Case 6. **A** Angiography of right external carotid artery. *Arrowhead* shows previous parietal EDAS. *Arrow* shows occipital EDAS. **B** Angiography of right internal carotid artery

veloped, and we consider this is because the simple EDAS procedures were applied.

Regarding the response to surgery, revascularization develops in around 70% of EDAS [15]. One explanation for the difference in revascularization is inhomogeneity in CBF between age and area [13]. We also obtained around 70% revascularization. However, this is the result of each EDAS. We combined frontal, parietal and occipital EDAS, so that all patients obtained some revascularization. With a combination of parietal EDAS, frontal EDAS and occipital EDAS, almost all cortical surface areas can be covered. If the development of revascularization is poor in one EDAS site, other EDAS sites can cover the low-perfusion areas, as in case 5. Y. Matsushima et al. reported that combined frontal and parietal EDAS is more effective than single EDAS [19]. Based on a similar idea, T. Matsushima et al. also recommended EMAS in the frontal lobe [16, 17]. A combination of parietal and frontal and/or occipital EDAS appears to offer adequate perfusion.

Regarding the timing of additional surgery, we used to wait more than 4 months, but we now perform additional surgery sooner. Simultaneous EDAS using the parietal branch and the frontal branch and/or the occipital artery may be effective in some patients as in case 7 [14, 19].

In conclusion, multiple EDAS using the frontal branch of STA and the occipital artery is an effective and safe method of preventing ischemia in the frontal and occipital lobe in pediatric moyamoya disease.

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