Comparison of Bifrontal Craniotomy and Multiple Burr Hole Encephalogaleoperiosteal-Synangiosis for Pediatric Moyamoya Disease: An Experience of 346 Patients

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Received, January 10, 2023; **Accepted,** February 23, 2023; **Published Online,** April 14, 2023.

BACKGROUND: Moyamoya disease (MMD) is a steno-occlusive disease treated with revascularization surgery. Craniotomy and multiple burr hole encephalogaleoperiosteal-synangiosis (EGPS) are used for revascularization of the anterior cerebral artery territory.

OBJECTIVE: To compare the clinical outcome between the 2 surgical methods in pediatric patients with MMD.

METHODS: A retrospective review of patients with MMD who underwent bifrontal indirect bypass surgery was performed. Clinical features, perioperative data, and angiographic, perfusion, and functional outcomes were compared between the 2 groups. Propensity score matching was performed to compare the perioperative characteristics and clinical outcomes.

RESULTS: A total of 346 patients were included in this study, 111 patients underwent bifrontal craniotomy EGPS, and 235 patients had bifrontal multiple burr hole EGPS. An insignificant higher rate of postoperative infarction (11.7% vs 5.5%, $P = .072$) and more postoperative hemorrhage occurred in the craniotomy EGPS group (3.6% vs 0%, $P = .004$). Of the 83 patients selected with propensity score matching for each group, the duration of operation was shorter ($P < .001$) and the amount of intraoperative bleeding was significantly less in the multiple burr hole EGPS group ($P = .008$). There was no difference in clinical outcomes between the 2 groups.

CONCLUSION: Bifrontal multiple burr hole EGPS has benefits over craniotomy with shorter surgical time, less intraoperative bleeding, fewer postoperative complications, and comparable perfusion and functional outcomes. Multiple burr hole EGPS is a safe and effective method that might be considered for revascularization of the anterior cerebral artery territory in pediatric patients with MMD.

KEY WORDS: Burr hole, Frontal, Indirect revascularization, Moyamoya disease, Pediatric

Neurosurgery 00:1–11, 2023 https://doi.org/10.1227/neu.0000000000002499

Moyamoya disease (MMD) is a steno-occlusive disease of the bilateral internal carotid arteries that predominantly occurs in East Asia.[1,2] In pediatric patients with MMD, revascularization through indirect bypass surgery is widely used and has proven satisfactory outcomes.[3,4] Because the middle cerebral artery (MCA) and anterior cerebral artery (ACA) are involved in most patients, efforts have been made to achieve revascularization of the frontal region, not only confined to the parietal region.[5,6]

Various methods of indirect bypass surgery are performed depending on the types of donor tissue, the size and location of the craniotomy, and the use of burr hole procedures.[7,8] For

ABBREVIATIONS: EDAS, encephaloduroarterio-synangiosis; **EFS,** event-free survival; **EGPS,** encephalogaleoperiosteal-synangiosis; **FSIQ,** full-scale intelligence quotient; **MMD,** moyamoya disease; **PCA,** posterior cerebral artery; **STA,** superficial temporal artery.

revascularization of the ACA territory, we have performed bifrontal craniotomy encephalogaleoperiosteal-synangiosis (EGPS) for decades.[9] In addition, a modified burr hole EGPS procedure similar to that described by Kawamoto et al[10] was used as an alternative to craniotomy in patients with preexisting transdural collaterals. Afterward, we gradually expanded our application of the bifrontal multiple burr hole EGPS for revascularization of the ACA territory.

Several studies have reported the feasibility of burr hole surgery in the ACA territory but are limited to a small number of patients, with no comparative data with craniotomy procedures.[11-13] Our clinical experience with a considerable number of pediatric patients with MMD, consistent treatment policy, and standardized surgical procedure provides well-organized comparative data of the bifrontal multiple burr hole EGPS with craniotomy EGPS. The goal of this study was to review the surgical outcome of bifrontal multiple burr hole EGPS in pediatric patients with MMD and extend the application of multiple burr hole EGPS for revascularization of the ACA territory. We have focused on the detailed operative technique and have emphasized minimizing perioperative complications in pediatric MMD surgery.

METHODS

Patient Selection

From June 2006 to December 2020, 395 patients underwent bifrontal indirect bypass surgery combined with unilateral superficial temporal artery (STA) encephaloduroarterio-synangiosis (EDAS) by a single surgeon (S-K. K.) at our institute. Twenty-eight patients with moyamoya syndrome and 21 patients with poor follow-up data were excluded from this study. Therefore, a total of 346 patients were included in this study. Patients were divided into 2 groups, according to the type of bifrontal indirect bypass surgery, and a propensity score matching was performed (Figure 1). Informed consent was waived. This study protocol was approved by the local Institutional Review Board (IRB No. 2211-099-1378) and was conducted according to the Declaration of Helsinki.

Surgical Techniques

All patients included in this study underwent bifrontal indirect bypass surgery (craniotomy EGPS, multiple burr hole EGPS) combined with unilateral STA EDAS simultaneously. In our institute, surgery for patients with MMD is usually performed in 2 stages, initially in the symptomatic and hemodynamically affected hemisphere. Bifrontal indirect bypass surgery was performed simultaneously with unilateral STA EDAS when symptoms involving the ACA territory were present or when insufficient ACA territory perfusion was seen in the perfusion study.

Bifrontal craniotomy EGPS was performed according to previous papers (Figure 2A).[9,14] A scalp incision was made anteriorly, parallel to the coronal suture with a curvilinear line. The galeoperiosteal flap was harvested, and a rectangular-shaped (4 × 8-cm sized) craniotomy crossing the midline was performed. Durotomy was performed sparing the superior sagittal sinus, and the dural flaps were reflected into the subdural space of the lateral frontal convexity. A galeoperiosteal flap was placed onto each side of the frontal area and sutured with the incised dura margin.

Between 2006 and 2015, bifrontal multiple burr hole EGPS was preferred over craniotomy EGPS when preexisting transdural collateral channels were present in the frontal area and in patients who had previous infarctions in the frontal area. Since 2016, bifrontal multiple burr hole EGPS has been implemented for all patients who need revascularization in the ACA territory due to our preference for a less invasive procedure in pediatric patients with MMD. A scalp incision was made curvilinearly in front of the coronal suture, similar to bifrontal craniotomy EGPS. The midline was marked, and 3 rectangular galeoperiosteal flaps were made obliquely on each side with their base away from the midline. To insert

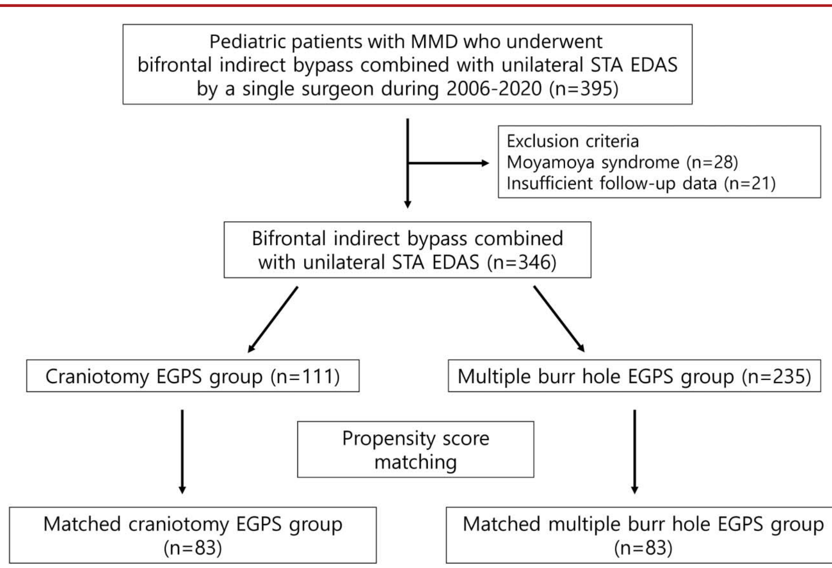

FIGURE 1. *Patient flowchart of this study. EDAS, encephaloduroarterio-synangiosis; EGPS, encephalogaleoperiosteal-synangiosis; MMD, moyamoya disease; STA, superficial temporal artery.*

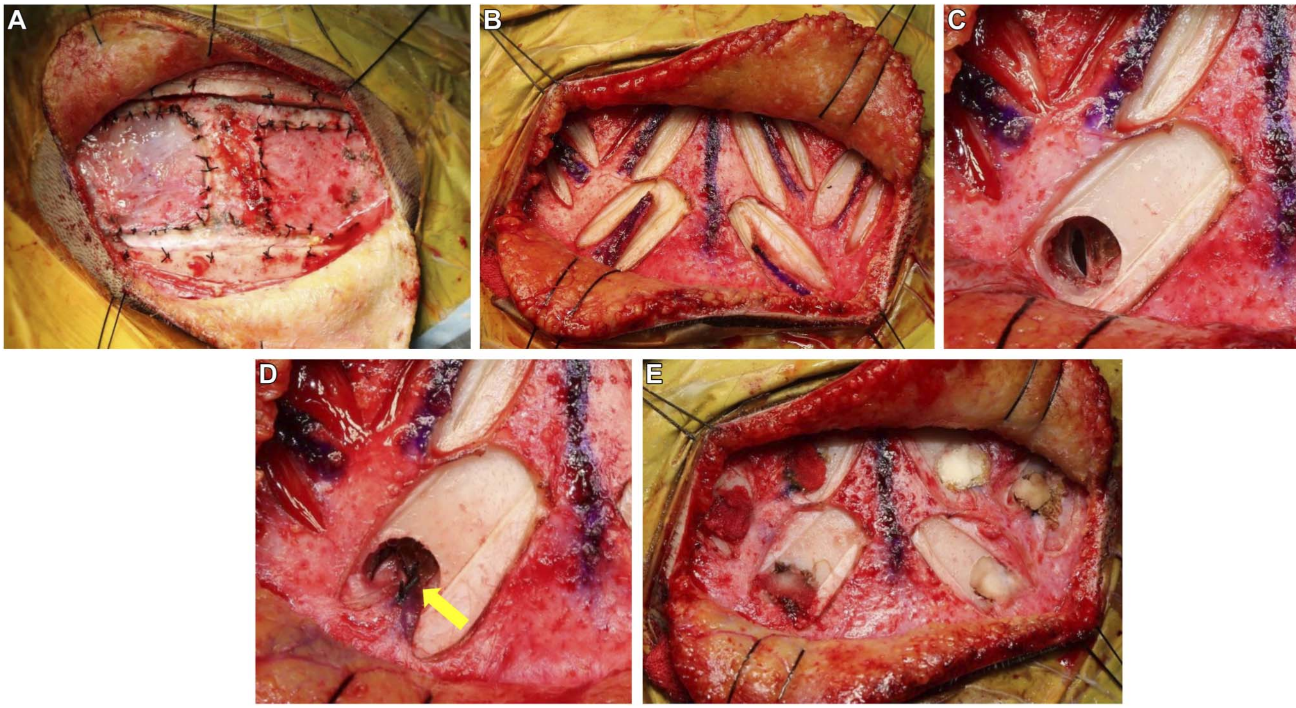

FIGURE 2. *Intraoperative photograph of surgical procedures.* **A**, *The bifrontal craniotomy EGPS was performed according to our previous reports.*[9,14] *The shape of galeoperiosteal flaps sutured onto each side of the dura margin and sparing the midline superior sagittal sinus.* **B**, *For the bifrontal multiple burr hole EGPS, 3 rectangular galeoperiosteal flaps were designed at each side with their base away from the midline. The long flap design involves inserting as many flaps as possible.* **C**, *Burr holes were made at the base of each flap, which is also intended to insert more tissue.* **D**, *The harvested galeoperiosteal flap was inserted into the brain surface and anchored to the dura margin with sutures (yellow arrow).* **E**, *A total of 6 burr hole EGPSs, 3 on each side, were made. Burr hole sites were reconstructed with pieces of TachoSil (Takeda) and SPONGOSTAN (Ethicon Inc). EGPS, encephalogaleoperiosteal-synangiosis.*

the maximum amount of flap possible, we designed long galeoperiosteal flaps (Figure 2B). After reflecting the flap, to avoid injury to the superior sagittal sinus, the most medial burr hole was made at a distance of approximately 1 cm from the midline, and then, based on this burr hole, separate burr holes were made on the lateral and posterior sides. Linear durotomy was performed, and the harvested galeoperiosteal flap was inserted into the brain surface by anchoring sutures to the dura (Figure 2C-2D). For reconstruction of the burr hole site, pieces of TachoSil (Takeda) and SPONGOSTAN (Ethicon Inc) were packed. A total of 6 burr hole EGPSs, 3 on each side, were made (Figure 2E).

Postoperatively, wound pain was controlled by local nerve block and patient-controlled analgesia for pain control. Laboratory abnormalities were strictly corrected with a goal of hemoglobin level >13 g/dL and an international normalized ratio of prothrombin time <1.20.[15] Sufficient hydration was supplied (1.5 times of maintenance volume) and tapered off according to the patient's status. When prolonged neurological symptom or neurological deterioration was present, diffusion MRI was performed to rule out postoperative infarction and hemorrhage.

Outcome Assessment and Statistical Analysis

The preoperative angiographic stage was classified according to the criteria by Suzuki et al.[16] The amount of intraoperative bleeding, duration of operation, presence of postoperative infarction, and hemorrhage were evaluated for perioperative outcomes. The postoperative angiographic outcome was not routinely evaluated due to the invasive nature of this study. Patients who had combined bifrontal indirect bypass as the first operation underwent angiographic evaluation 2 to 6 months postoperatively before the second operation. The angiographic outcome of the ACA territory was classified into 3 categories modified from the grading system by Matsushima et al[17]: *A* indicates collateral formation in more than two-thirds of the ACA distribution; *B* indicates collateral formation in less than two-thirds of the ACA distribution; and *C* indicates faint or no visible collateral formation (Figure 3A). The perfusion outcome was classified into 2 categories: improved and no change/worsened according to the time to peak images on follow-up perfusion MRI taken every 1 year until the second year after the operation and every 2 years until the sixth year after the operation (Figure 3B).[18] Cognitive function was evaluated with the full-scale intelligence quotient (FSIQ) 1 year postoperatively. The cut-off value for a favorable cognitive outcome was 90. The neurological outcome was evaluated using the modified Rankin Scale (mRS) score.

To compare clinical characteristics between the 2 surgery groups, a χ^2 test was used for discrete variables and the Student *t*-test was used for continuous variables. The amount of intraoperative bleeding and duration of operation were compared between the multiple burr hole EGPS and bifrontal EGPS groups. Intraoperative bleeding was measured and recorded

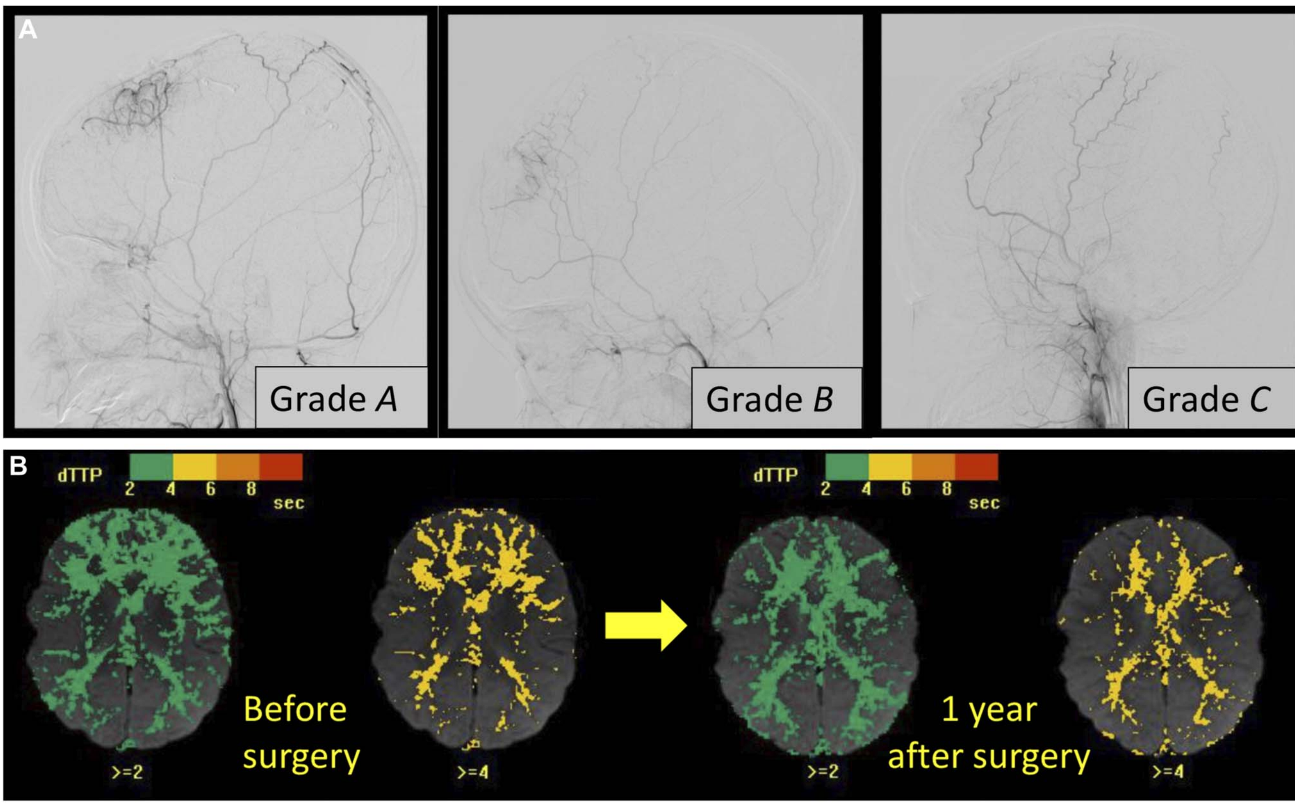

FIGURE 3. *The classification of revascularization outcomes.* **A**, *Angiographic classification of the ACA territory modified from the grading system by Matsushima et al.*[17] *A indicates collateral formation in more than two-thirds of the ACA distribution; B indicates collateral formation in less than two-thirds of the ACA distribution; and C indicates faint or no visible collateral formation.* **B**, *Perfusion outcome is classified into 2 categories: improved and no change/worsened according to the time to peak images on follow-up perfusion MRI. The MRI of a 9-year-old boy shows improvement of time to peak delay in the frontal region at 1-year postoperative follow-up imaging. ACA, anterior cerebral artery.*

by anesthesiologists. The duration of operation and intraoperative blood loss can be affected by factors such as age, body weight due to thickness of the calvarium, and development of vessels in the scalp and dura layer, and performing arachnoid dissection during the STA EDAS procedure has substantial effects on operation time.[19] To eliminate these confounding factors, propensity score matching was performed between the 2 groups with a standardized mean difference of 0.25. A Kaplan-Meier curve was drawn for event-free survival (EFS) of ischemic or hemorrhagic infarction occurrence during the follow-up. Immediate postoperative infarction and hemorrhages were excluded from the analysis of EFS.

RESULTS

Patient Characteristics

Among the 346 patients included in this study, 111 had received bifrontal craniotomy EGPS combined with STA EDAS and 235 had received bifrontal multiple burr hole EGPS combined with STA EDAS. There was no significant sex difference between the 2 groups. The median ages were 8.2 years and 7.7 years, respectively. Preoperative characteristics showed a significant difference between the 2 groups. Preoperative infarction was more common in the multiple burr hole EGPS group (P = .013) in both cortical/watershed infarct (craniotomy EGPS 15.3% vs multiple burr hole EGPS 24.3%) and large territorial infarct (craniotomy EGPS 0.9% vs multiple burr hole EGPS 5.5%). As a consequence, the proportion of poor graders in the preoperative mRS score was significantly higher in the multiple burr hole EGPS group than in the craniotomy EGPS group (P = .014). In addition, as we mentioned earlier, because multiple burr hole EGPS was preferred when transdural collateral was present in the ACA territory, the preoperative angiographic stage differed between the 2 groups. The rate of additional surgery due to posterior cerebral artery (PCA) involvement was significantly higher in the multiple burr hole EGPS group (craniotomy EGPS 13.5% vs multiple burr hole EGPS 27.2%, P = .007). As multiple burr hole EGPS has been performed in all patients since 2016, the follow-up period was significantly different between the 2 groups (Table 1).

TABLE 1. Clinical Characteristics of the Patients

Clinical features	Craniotomy EGPS group (n = 111)	Multiple burr hole EGPS group (n = 235)	P value
	Number of cases (%) or value		
Sex			.177
M	58 (52.3%)	103 (43.8%)	
F	53 (47.7%)	132 (56.2%)	
Age (y)	8.2 ± 3.4	7.7 ± 3.6	.281
Weight (kg)	33.0 ± 14.1	33.0 ± 15.9	.981
Height (cm)	130.3 ± 20.0	128.7 ± 22.1	.511
Preoperative mRS score			.014
0-2	106 (95.5%)	202 (86.0%)	
3-5	5 (4.5%)	33 (14.0%)	
Preoperative infarction			.013
None	93 (83.8%)	165 (70.2%)	
Cortical/watershed area	17 (15.3%)	57 (24.3%)	
Large territory	1 (0.9%)	13 (5.5%)	
Angiographic stages[a] (n = 344)	110	234	<.001
I-III	85 (78.0%)	129 (55.1%)	
IV-VI	24 (22.0%)	105 (44.9%)	
Surgery due to PCA involvement during follow-up			0.007
None	96 (86.5%)	171 (72.8%)	
Performed	15 (13.5%)	64 (27.2%)	
Follow-up period (mo)	107.8 ± 45.2	63.5 ± 40.0	<.001

EGPS, encephalogaleoperiosteal-synangiosis; mRS, modified Rankin Scale; PCA, posterior cerebral artery.
[a]Angiographic stages were evaluated according to the criteria by Suzuki et al.[16]

Perioperative Data and Postoperative Complications

The incidence of postoperative infarction was not significantly different, but a higher rate of infarction occurred in the craniotomy EGPS group (craniotomy EGPS 11.7% vs multiple burr hole EGPS 5.5%, P = .072). The incidence of postoperative hemorrhages showed a significant difference as none occurred in the multiple burr hole EGPS group (craniotomy EGPS 3.6% vs multiple burr hole EGPS 0%, P = .004). The patient status that required reoperation was not different between the 2 groups, and all reoperations were performed at the STA EDAS site (Table 2).

Revascularization and Clinical Outcomes

The angiographic outcome was significantly different between the 2 groups, with a higher rate of grade A revascularization in the craniotomy EGPS group (P = .008); however, considering grades A and B together showed no significant difference (craniotomy EGPS 98% vs multiple burr hole EGPS 94%). The perfusion outcome with follow-up perfusion MRI showed no difference in the proportion of improved results between the 2 groups (Table 3).

Over 70% of the patients were symptom-free at the last follow-up, and headaches were the most common residual symptom in both groups (craniotomy EGPS 13.5% vs multiple burr hole EGPS 20.8%), followed by transient ischemic symptoms (craniotomy EGPS 9.9% vs multiple burr hole EGPS 6.4%). More than 85% of patients with headaches had a frequency of once a week or less, and they were either self-limiting or well controlled with medication. The cognitive function outcome was not significantly different between the 2 groups, with a favorable outcome rate of 75.6% in the craniotomy EGPS group and 66.3% in the multiple burr hole EGPS group.

The change of mRS score at the last follow-up showed improved results for all patients (Figure 4). There were only 2

TABLE 2. Perioperative Characteristics of the Patients

Perioperative data	Craniotomy EGPS group (n = 111)	Multiple burr hole EGPS group (n = 235)	P value
	Number of cases (%) or value		
Arachnoid dissection at STA EDAS site			<.001
Performed	104 (93.7%)	83 (35.3%)	
Preserved	7 (6.3%)	152 (64.7%)	
Duration of operation (min)	356.7 ± 53.2	278.3 ± 48.1	<.001
Intraoperative blood loss (cc)	458.7 ± 276.2	287.6 ± 188.3	<.001
Postoperative infarction			.072
None	96 (86.5%)	212 (90.2%)	
Present	13 (11.7%)	13 (5.5%)	
STA EDAS site	2 (1.8%)	10 (4.3%)	
Postoperative hemorrhage			.004
None	102 (91.9%)	231 (98.3%)	
Present	4 (3.6%)	0 (0.0%)	
STA EDAS site	5 (4.5%)	4 (1.7%)	
Reoperation due to postoperative complications			.305
None	107 (96.4%)	232 (98.7%)	
Performed[a]	4 (3.6%)	3 (1.3%)	

EDAS: encephaloduroarterio-synangiosis; EGPS, encephalogaleoperiosteal-synangiosis; STA, superficial temporal artery.
[a]All reoperations were performed at STA EDAS site.

and 3 patients who developed infarction or hemorrhage, respectively, during follow-up, and the Kaplan-Meier curve for EFS and a log-rank test showed no significant difference between the groups (Figure 5). The 5-year EFS was 98.2% for the craniotomy EGPS group and 99.1% for the multiple burr hole EGPS group. The 10-year EFS was 98.2% for the craniotomy EGPS group and 98.0% for the multiple burr hole EGPS group.

Propensity Score Matching for Perioperative Characteristics and Clinical Outcomes

Propensity score matching was performed for the 2 groups with age, weight, preoperative mRS score, and arachnoid dissection. As a result, 83 patients were selected for each group. The duration of operation was significantly shorter in the multiple burr hole EGPS group (craniotomy EGPS 354.2 ± 52.2 vs multiple burr hole EGPS 308.6 ± 49.5 minutes, P < .001), and the amount of intraoperative blood loss was less in the multiple burr hole EGPS group (craniotomy EGPS 437.5 ± 213.2 vs multiple burr hole EGPS 343.7 ± 229.0 cc, P = .008). The postoperative complications and clinical outcomes were not significantly different between the 2 groups (Table 4).

DISCUSSION

Indirect revascularization using the burr hole procedure for MMD has been reported in several studies since it was first described by Endo et al.[11] First, burr hole procedures described in the literature are quite different. Burr hole procedures were either performed by opening the dura without inserting any tissue[11,13,20] or inserting a harvested tissue flap in the burr hole site to make contact with the brain.[10,21,22] The multiple burr hole EGPS procedure described in this study also inserts galeoperiosteal flaps into the intradural space.

Revascularization of the ACA territory improves clinical outcomes, including cognitive function and angiographic outcomes in the ACA territory.[6,23] We have previously reported a study suggesting the favorable outcomes of bifrontal craniotomy EGPS combined with STA EDAS compared with STA EDAS alone.[9] We have also performed a bifrontal multiple burr hole EGPS procedure for patients who had preexisting transdural collaterals or previous infarctions in the frontal area as it is important not to disrupt preexisting collaterals and less invasive surgery was preferred.[20,24] Recently, we have favored less invasive procedures in pediatric patients with MMD. Therefore, since 2016, bifrontal multiple burr hole EGPS has been performed rather than bifrontal craniotomy EGPS for patients requiring revascularization in the ACA territory.

TABLE 3. Revascularization and Clinical Outcomes of the Patients

Outcomes	Craniotomy EGPS group (n = 111)	Multiple burr hole EGPS group (n = 235)	*P* value
	Number of cases (%) or value		
Angiographic outcome (n = 165)[a]	49	116	.008
A	44 (89.8%)	77 (66.4%)	
B	4 (8.2%)	32 (27.6%)	
C	1 (2.0%)	7 (6.0%)	
Perfusion outcome			.944
Improved	73 (65.8%)	157 (66.8%)	
No change/worsened	38 (34.2%)	78 (33.2%)	
Persistent symptoms			.336
Symptom free	82 (73.9%)	167 (71.1%)	
Transient ischemic attack	11 (9.9%)	15 (6.4%)	
Seizure	3 (2.7%)	7 (3.0%)	
Headache	15 (13.5%)	49 (20.8%)	
Once a week or less	13	44	
More than once a week	2	5	
Cognitive function outcome (n = 271)	90	181	.156
FSIQ ≥90	68 (75.6%)	120 (66.3%)	
FSIQ <90	22 (24.4%)	61 (33.7%)	

EGPS, encephalogaleoperiosteal-synangiosis; FSIQ, full sequence intelligence quotient.
[a]Angiographic outcome was evaluated with modified grading system by Matsushima et al.[17]

The results of this study show that bifrontal multiple burr hole EGPS has advantages over bifrontal craniotomy EGPS regarding the duration of operation, amount of intraoperative bleeding, and postoperative complications, especially hemorrhage. Notably, no postoperative hemorrhage occurred in multiple burr hole EGPS patients, while a few cases still occurred in craniotomy EGPS patients. The burr hole procedure is less invasive and has fewer bleeding complications than the craniotomy procedure because of the smaller area of dural surface exposure and epidural detachment. In addition, a smaller area of the galeoperiosteal flap, which is a potential bleeding source, may contribute to this outcome. The results of this study have advantages because multiple burr hole EGPS is less invasive than craniotomy EGPS in a large number of pediatric patients with MMD.

Bifrontal multiple burr hole EGPS was also not inferior to craniotomy EGPS in clinical outcomes. There were only very few patients in the 2 groups who newly developed infarction or hemorrhage in the anterior circulation territory during long-term follow-up, and a favorable neurological outcome was seen with the follow-up mRS score. Although the perfusion outcome was not different between the 2 groups, the angiographic outcome showed significantly favorable results in the craniotomy EGPS group. A wider surgical area shows better revascularization outcomes.[23] Another explanation for this result is that more previously infarcted patients were in the multiple burr hole EGPS group, and indirect bypass surgery does not show effective revascularization at the surface of the infarcted area.[20] More importantly, the rate of grade C revascularization was both very low and not different between the groups, and the assessment of revascularization through simple digital subtraction angiography can be subjective.[25] Regarding the symptomatic outcome, headaches were present in a considerable number of patients in our cohort. Headaches can persist regardless of the favorable revascularization outcome in anterior circulation.[26] Most headaches in our cohort were either self-limiting or well controlled with medication. However, PCA insufficiency can also cause headaches.[27,28] In these cases, revascularization of the PCA territory can be helpful as we have performed in our cohort. The higher rate of PCA involvement in the multiple burr hole EGPS group can also be related to the higher presence of infarction at the time of diagnosis.[29]

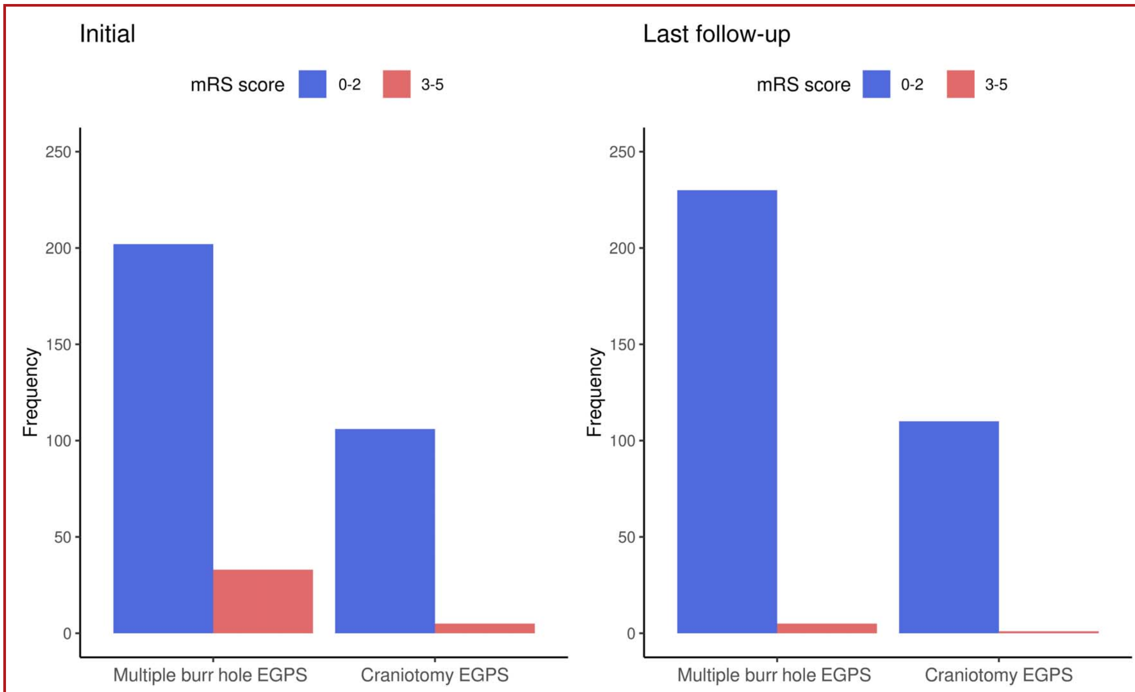

FIGURE 4. *The change in modified Rankin Scale (mRS) score shows the improvement in neurological outcomes in pediatric patients with moyamoya disease who had bifrontal multiple burr hole EGPS and craniotomy EGPS. Patients with favorable neurological status (mRS score 0-2) changed from 202/235 (85.9%) preoperatively to 230/235 (97.9%) at the last follow-up in the multiple burr hole EGPS group. In the craniotomy EGPS group, patients with favorable neurological status changed from 106/111 (95.5%) preoperatively to 110/111 (99.1%) at the last follow-up. Although 33 patients had poor mRS scores (3-5) in the multiple burr hole EGPS group, 28 of them improved to a favorable status. EGPS, encephalogaleoperiosteal-synangiosis; mRS, modified Rankin Scale.*

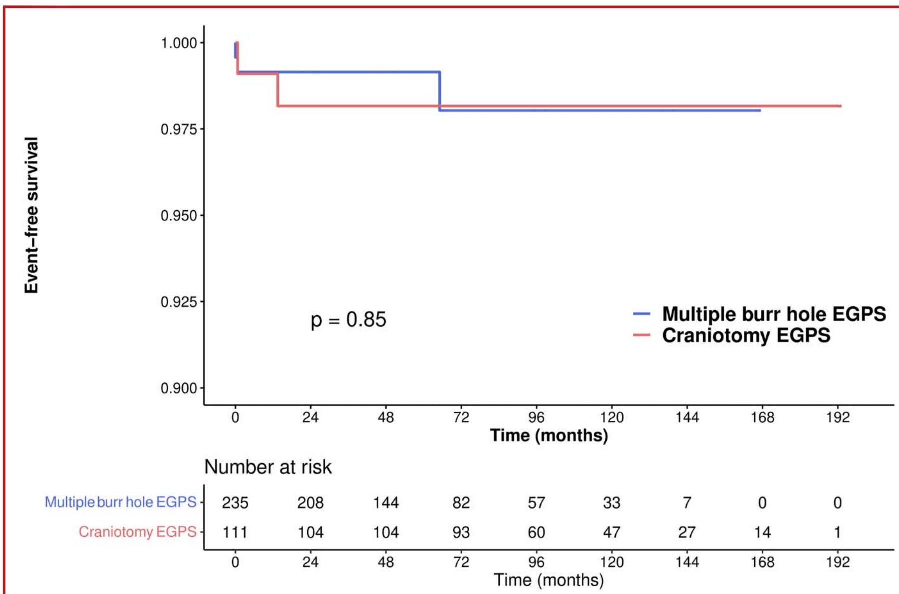

FIGURE 5. *The Kaplan-Meier curve for ischemic and hemorrhagic event occurrence and a log-rank test showed no significant difference between the craniotomy EGPS group and the multiple burr hole EGPS group (P = .85). The 5-year event-free survival was 98.2% for the craniotomy EGPS group and 99.1% for the multiple burr hole EGPS group. The 10-year EFS was 98.2% for the craniotomy EGPS group and 98.0% for the multiple burr hole EGPS group. EGPS, encephalogaleoperiosteal-synangiosis.*

TABLE 4. Propensity Score-Matched Data for Perioperative Characteristics and Clinical Outcomes

Clinical features and outcomes	Craniotomy EGPS group (n = 83)	Multiple burr hole EGPS group (n = 83)	*P* value
	Number of cases (%) or value		
Sex			.120
M	44 (53.0%)	33 (39.8%)	
F	39 (47.0%)	50 (60.2%)	
Age (y)	7.7 ± 3.4	8.1 ± 3.7	.420
Weight (kg)	28.6 ± 11.2	33.7 ± 16.3	.020
Preoperative mRS score			1.000
0-2	78 (94.0%)	78 (94.0%)	
3-5	5 (6.0%)	5 (14.0%)	
Arachnoid dissection at STA EDAS site			1.000
Performed	76 (91.6%)	76 (91.6%)	
Preserved	7 (8.4%)	7 (8.4%)	
Duration of operation (min)	354.2 ± 52.2	308.6 ± 49.5	<.001
Intraoperative blood loss (cc)	437.5 ± 213.2	343.7 ± 229.0	.008
Postoperative infarction			.114
None	75 (90.4%)	76 (91.6%)	
Present	8 (9.6%)	4 (4.8%)	
STA EDAS site	0 (0.0%)	3 (3.6%)	
Postoperative hemorrhage			.197
None	76 (91.6%)	80 (96.4%)	
Present	3 (3.6%)	0 (0.0%)	
STA EDAS site	4 (4.8%)	3 (3.6%)	
Angiographic outcome (n = 83)[a]	40	43	.385
A	36 (90.0%)	37 (86.0%)	
B	3 (7.5%)	6 (14.0%)	
C	1 (2.5%)	0 (0.0%)	
Perfusion outcome			1.000
Improved	52 (62.7%)	58 (69.9%)	
No change/worsened	31 (37.3%)	25 (30.1%)	
Cognitive function outcome (n = 141)	70	71	.492
FSIQ ≥90	52 (74.3%)	48 (67.6%)	
FSIQ <90	18 (25.7%)	23 (32.4%)	

EDAS, encephaloduroarterio-synangiosis; EGPS, encephalogaleoperiosteal-synangiosis; FSIQ, full sequence intelligence quotient; mRS, modified Rankin Scale; STA, superficial temporal artery.
[a]Angiographic outcome was evaluated with modified grading system by Matsushima et al.[17]

Efforts have been made to improve clinical and angiographic outcomes by making wider areas of revascularization surgery and using various types of tissues.[7,23,30] We also performed a wide craniotomy during the STA EDAS procedure on the MCA territory. However, burr hole procedures have shown promising results for the ACA territory.[13] Consistent with what Suzuki et al[12] claimed in their work, we agree that reducing perioperative complications and performing a less invasive procedure are important in patients with MMD. Basically, MMD surgery is a preventive operation against future cerebral infarction and hemorrhage. In addition, the long-term prognosis of children with MMD is closely related to the occurrence of perioperative complications.[3] Therefore, efforts should be made to achieve the same clinical outcome with less invasive surgery in pediatric MMD.

Recently, our clinical interest has been in reducing perioperative complications of pediatric MMD surgery. Intraoperative management to maintain normocapnia and adequate cerebral perfusion is of utmost importance. In our studies, arachnoid preservation,[19] avoiding double craniotomies (STA EDAS combined with craniotomy EGPS),[15,31] and subcutaneous drain insertion[31] were related to fewer postoperative infarctions and hemorrhagic complications. In addition, from this study, we provide the advantages of multiple burr hole EGPS and suggest that multiple burr hole EGPS is not just an alternative method to craniotomy EGPS but a safe and effective procedure that might be considered as the first option.

Limitations

There are some limitations of this study due to the retrospective nature of the study. In addition, as it is a single-center study, selection bias cannot be avoided. However, our results show satisfactory long-term follow-up data of the cohort. Furthermore, we suggest that a single surgeon's consistent treatment policy and surgical technique can provide better comparative data of surgical outcomes.

CONCLUSION

Bifrontal multiple burr hole EGPS for pediatric patients with MMD resulted in favorable revascularization and clinical outcomes. Moreover, multiple burr hole EGPS in the bifrontal area is a less invasive procedure with minimal operative bleeding and shorter operation time compared with craniotomy EGPS. The multiple burr hole EGPS may be the preferred method for revascularization of the ACA territory.

Funding

This research was supported and funded by the Seoul National University Hospital Kun-hee Lee Child Cancer & Rare Disease Project, Republic of Korea (Grant No. 70-2023-3111).

Disclosures

The authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices described in this article.

REFERENCES

1. Kuriyama S, Kusaka Y, Fujimura M, et al. Prevalence and clinicoepidemiological features of moyamoya disease in Japan: findings from a nationwide epidemiological survey. *Stroke*. 2008;39(1):42-47.
2. Kim SK, Cho BK, Phi JH, et al. Pediatric moyamoya disease: an analysis of 410 consecutive cases. *Ann Neurol*. 2010;68(1):92-101.
3. Ha EJ, Kim KH, Wang KC, et al. Long-term outcomes of indirect bypass for 629 children with moyamoya disease: longitudinal and cross-sectional analysis. *Stroke*. 2019;50(11):3177-3183.
4. Kuroda S, Houkin K. Moyamoya disease: current concepts and future perspectives. *Lancet Neurol*. 2008;7(11):1056-1066.
5. Matsushima T, Inoue T, Katsuta T, et al. An indirect revascularization method in the surgical treatment of moyamoya disease--various kinds of indirect procedures and a multiple combined indirect procedure. *Neurol Med Chir*. 1998;38(suppl):297-302.
6. Takahashi A, Kamiyama H, Houkin K, Abe H. Surgical treatment of childhood moyamoya disease. *Neurol Med Chir*. 1995;35(4):231-237.
7. Reis CVC, Safavi-Abbasi S, Zabramski JM, Gusmão SNS, Spetzler RF, Preul MC. The history of neurosurgical procedures for moyamoya disease. *Neurosurg Focus*. 2006;20(6):1-5.
8. Houkin K, Kuroda S, Ishikawa T, Abe H. Neovascularization (angiogenesis) after revascularization in moyamoya disease. Which technique is most useful for moyamoya disease? *Acta Neurochir*. 2000;142(3):269-276.
9. Kim SK, Wang KC, Kim IO, Lee DS, Cho BK. Combined encephaloduroarteriosynangiosis and bifrontal encephalogaleo(periosteal)synangiosis in pediatric moyamoya disease. *Neurosurgery*. 2002;50(1):88-96.
10. Kawamoto H, Kiya K, Mizoue T, Ohbayashi N. A modified burr-hole method 'galeoduroencephalosynangiosis' in a young child with moyamoya disease. *Pediatr Neurosurg*. 2000;32(5):272-275.
11. Endo M, Kawano N, Miyasaka Y, Yada K. Cranial burr hole for revascularization in moyamoya disease. *J Neurosurg*. 1989;71(2):180-185.
12. Suzuki Y, Negoro M, Shibuya M, Yoshida J, Negoro T, Watanabe K. Surgical treatment for pediatric moyamoya disease: use of the superficial temporal artery for both areas supplied by the anterior and middle cerebral arteries. *Neurosurgery*. 1997;40(2):324-330; discussion 329-330.
13. Dusick JR, Gonzalez NR, Martin NA. Clinical and angiographic outcomes from indirect revascularization surgery for moyamoya disease in adults and children: a review of 63 procedures. *Neurosurgery*. 2011;68(1):34-43; discussion 43.
14. Park JH, Yang SY, Chung YN, et al. Modified encephaloduroarteriosynangiosis with bifrontal encephalogaleoperiosteal synangiosis for the treatment of pediatric moyamoya disease. *J Neurosurg Pediatr*. 2007;106(3):237-242.
15. Choi JW, Chong S, Phi JH, et al. Postoperative symptomatic cerebral infarction in pediatric moyamoya disease: risk factors and clinical outcome. *World Neurosurg*. 2020;136:e158-e164.
16. Suzuki J, Kodama N. Moyamoya disease--a review. *Stroke*. 1983;14(1):104-109.
17. Matsushima T, Inoue T, Suzuki SO, Fujii K, Fukui M, Hasuo K. Surgical treatment of moyamoya disease in pediatric patients--comparison between the results of indirect and direct revascularization procedures. *Neurosurgery*. 1992;31(3):401-405.
18. Kim SK, Wang KC, Oh CW, et al. Evaluation of cerebral hemodynamics with perfusion MRI in childhood moyamoya disease. *Pediatr Neurosurg*. 2003;38(2):68-75.
19. Byun YH, Phi JH, Lee JY, et al. Preservation of the arachnoid membrane during encephaloduroarteriosynangiosis reduces postoperative complications without undermining the surgical outcome in pediatric moyamoya disease. *World Neurosurg*. 2019;130:e406-e416.
20. Kawaguchi T, Fujita S, Hosoda K, et al. Multiple burr-hole operation for adult moyamoya disease. *J Neurosurg*. 1996;84(3):468-476.
21. Blauwblomme T, Mathon B, Naggara O, et al. Long-term outcome after multiple burr hole surgery in children with moyamoya angiopathy: a single-center experience in 108 hemispheres. *Neurosurgery*. 2017;80(6):950-956.
22. Sainte-Rose C, Oliveira R, Puget S, et al. Multiple bur hole surgery for the treatment of moyamoya disease in children. *J Neurosurg Pediatr*. 2006;105(6):437-443.
23. Kuroda S, Houkin K, Ishikawa T, et al. Determinants of intellectual outcome after surgical revascularization in pediatric moyamoya disease: a multivariate analysis. *Childs Nervous Syst*. 2004;20(5):302-308.
24. McLaughlin N, Martin NA. Effectiveness of burr holes for indirect revascularization in patients with moyamoya disease-a review of the literature. *World Neurosurg*. 2014;81(1):91-98.

25. Cho HH, Cheon JE, Kim SK, et al. Quantitative assessment of neovascularization after indirect bypass surgery: color-coded digital subtraction angiography in pediatric moyamoya disease. *Am J Neuroradiol*. 2016;37(5):932-938.
26. Seol HJ, Wang KC, Kim SK, Hwang YS, Kim KJ, Cho BK. Headache in pediatric moyamoya disease: review of 204 consecutive cases. *J Neurosurg Pediatr*. 2005;103(5):439-442.
27. Lee JY, Choi YH, Cheon JE, et al. Delayed posterior circulation insufficiency in pediatric moyamoya disease. *J Neurol*. 2014;261(12):2305-2313.
28. Lee JY, Kim S-K, Phi JH, Wang K-C. Posterior cerebral artery insufficiency in pediatric moyamoya disease. *J Korean Neurosurg Soc*. 2015;57(6):436-439.
29. Lee JY, Kim SK, Cheon JE, et al. Posterior cerebral artery involvement in moyamoya disease: initial infarction and angle between PCA and basilar artery. *Childs Nerv Syst*. 2013;29(12):2263-2269.
30. Fiaschi P, Scala M, Piatelli G, et al. Limits and pitfalls of indirect revascularization in moyamoya disease and syndrome. *Neurosurg Rev*. 2021;44(4):1877-1887.
31. Choi H, Lee JY, Phi JH, Kim SK, Cho BK, Wang KC. Postoperative epidural hematoma covering the galeal flap in pediatric patients with moyamoya disease: clinical manifestation, risk factors, and outcomes. *J Neurosurg Pediatr*. 2013;12(2):181-186.