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Anatomic Variation of the Superficial Temporal Artery and Posterior Auricular Artery in a Pediatric Moyamoya Disease Population

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ABSTRACT

BACKGROUND AND PURPOSE: In certain cases of pediatric patients with Moyamoya disease undergoing encephaloduroarteriosynangiosis (EDAS) treatment, the posterior auricular artery can be used as an alternative when the parietal branch of the superficial temporal artery is unavailable. In this study, anatomic variations of the superficial temporal and posterior auricular arteries in pediatric patients with Moyamoya disease and postoperative outcomes of posterior auricular artery-EDAS are explored.

MATERIALS AND METHODS: Medical records of 572 patients with Moyamoya disease who underwent surgical procedures from 2007 to 2017 at the Seoul National University Children's Hospital were reviewed. Anatomic classifications of the superficial temporal and posterior auricular arteries were based on previous classifications. Postoperative hemodynamic changes of posterior auricular artery-EDAS were analyzed using the Matsushima grade. Also, Karnofsky Performance Scale and mRS scores of posterior auricular artery-EDAS cases were reviewed to identify postoperative clinical outcomes.

RESULTS: Among 1144 hemispheres, 24 were considered posterior auricular artery-EDAS candidates (2.1%). Of those, 10 hemispheres underwent posterior auricular artery-EDAS (41.7%, in total hemispheres 0.9%). Comparing the Matsushima grades of the superficial temporal artery-EDAS and posterior auricular artery-EDAS groups showed similar postoperative revascularization. Postoperative Karnofsky Performance Scale and mRS scores of patients having undergone posterior auricular artery-EDAS did not show deterioration.

CONCLUSIONS: In approximately 2% of pediatric patients with Moyamoya disease for whom the superficial temporal artery is unavailable as the EDAS donor, the posterior auricular artery can be considered an alternative. On the basis of the results, the clinical outcome of posterior auricular artery-EDAS was not inferior to that of superficial temporal artery-EDAS. Hence, we suggest an in-depth consideration of the posterior auricular artery as the donor artery if the superficial temporal artery parietal branch is unavailable.

ABBREVIATIONS: EDAS = encephaloduroarteriosynangiosis; EGS = encephalogaleosynangiosis; MMD = Moyamoya disease; PAA = posterior auricular artery; STA = superficial temporal artery

Moyamoya disease (MMD) is one of the most common pediatric cerebrovascular diseases in East Asia, characterized by slowly progressive occlusion of the internal carotid arteries or their branches. Such progression causes the development of extensive collateral vessels known as Moyamoya vessels at the base of the brain.[1,2]

Among various symptoms, repeated TIAs are most prevalent in pediatric patients.[3] To increase the blood supply in MMD, direct or indirect bypass surgery is performed to enhance collateral revascularizations to the ischemic areas of the brain. Traditionally, due to technical difficulties involving the small size of donor and recipient vessels, indirect extracranial-intracranial bypass has been favored for pediatric patients with MMD.[4-8] Among various indirect bypass techniques, encephaloduroarteriosynangiosis (EDAS) is commonly performed to increase the blood supply to the MCA territory. Here, the parietal branch of the superficial temporal artery (STA) is commonly used as the donor artery, given its proximity to the motor cortex. However, the parietal branch of the STA is sometimes unavailable, as with a previously used STA parietal branch and hypoplasia, absence, or anterior positioning of the STA parietal branch. Then, the posterior auricular artery (PAA) with the appropriate anatomic criteria met could be used as an alternative.

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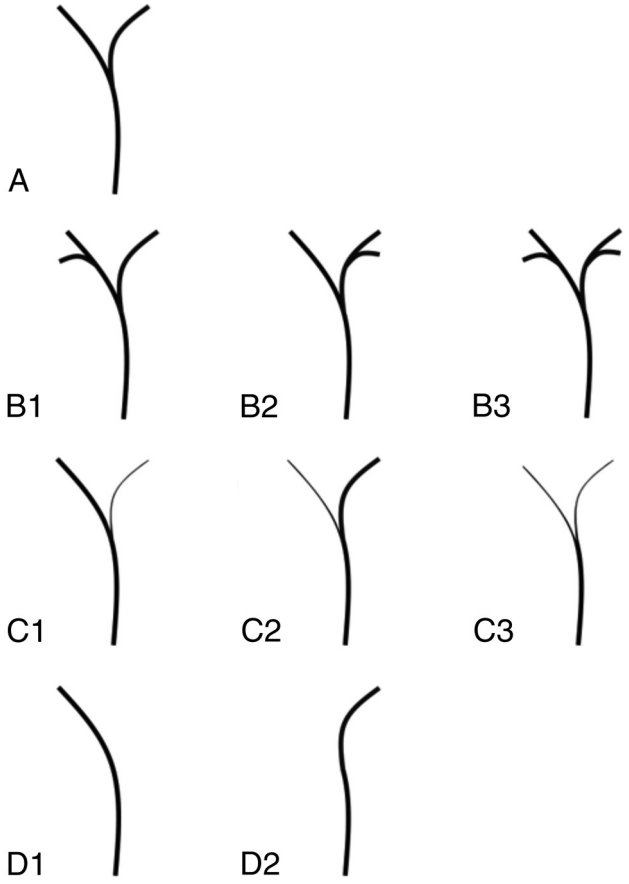

FIG 1. STA classification. Class A corresponds to classic STA anatomy with a single frontal and parietal branch. Classes B1, B2, and B3 illustrate additional frontal, parietal, or both branches, respectively. Class C1, C2, and C3 illustrate hypoplasia of the parietal, frontal, or both branches, respectively. Class D1 and D2 illustrate aplasia of the parietal and frontal branches, respectively.

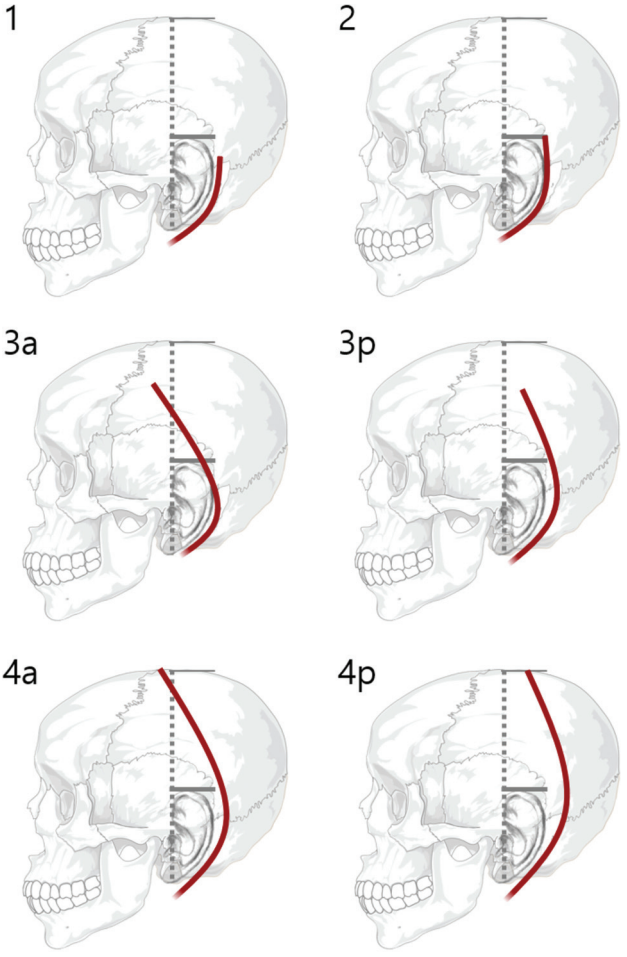

FIG 2. PAA classification. The *thin solid line* indicates the vertex; the *dashed vertical line* indicates the external auditory canal midline; and the *thick solid line* indicates the helix. Class 1 illustrates PAA terminating below the helix, class 2 illustrates the PAA terminating at the helix, class 3 illustrates the PAA terminating between the helix and the vertex, and class 4 illustrates the PAA terminating at the vertex. For classes 3 and 4, if the PAA advances anteriorly passing the vertical line, the class is annotated with "a" for anteriority and, if not, "p" for posteriority.

There are several reports in the literature of successful use of the PAA as an alternative.[9–12] Yet, no research has been conducted on PAA use as an indirect bypass alternative, especially in pediatric patients. Also, there has been no thorough investigation of STA and PAA anatomic variations on a large number of pediatric patients with MMD.

In this article, we investigate the anatomic variations of both the STA and PAA in pediatric patients with MMD and examine the STA and PAA anatomic variation combinations that are considered adequate for performing PAA-EDAS. Additionally, postoperative clinical outcomes and the degree of postoperative revascularization of STA-EDAS and PAA-EDAS are compared.

MATERIALS AND METHODS

From January 1, 2007, to December 31, 2017, we surgically treated 600 patients with MMD. Of those, 572 patients with MMD were enrolled in this study after using the following exclusion criteria: 12 patients with Moyamoya syndrome and 16 with insufficient electronic medical record data or radiologic records. To classify the STA, we initially considered adopting the STA classification coined by Marano et al.[13] However, due to excessive subgroups and complexity, as illustrated in Fig 1, we opted for the classification by Medved et al[14] without class E which illustrates an additional superior auricular artery, which is not crucial to surgical planning.

Likewise, we adopted the PAA classification coined by Tokugawa et al[15] with some modifications. The alphabetic classification was changed to numeric for easier distinction between STA and PAA classes, as well as the anteriority and posteriority of classes 3 and 4 as illustrated in Fig 2.

To evaluate the postoperative technical outcomes of PAA-EDAS and STA-EDAS, we compared the degree of revascularization on follow-up transfemoral cerebral angiography between the PAA-EDAS and STA-EDAS groups based on the Matsushima grade: Grade A denotes revascularization of more than two-thirds of the MCA territory, grade B denotes revascularization of between one- and two-thirds of the MCA territory, and grade C denotes revascularization of a slight amount or none of the territory.[16] Twice as many patients having undergone STA-EDAS ($n = 20$) as those in the PAA-EDAS group ($n = 10$) were selected for the same age-

Table 1: Demographics of 572 patients

Demographics	Value
Sex ($n = 572$)	
Male	255 (44.6%)
Female	317 (55.4%)
Age of diagnosis (yr)	
Younger than 3	55 (9.7%)
3∼7	230 (40.2%)
8∼11	188 (32.8%)
12∼14	73 (12.7%)
15∼18	26 (4.5%)
Unilateral ($n = 112$)	
Right	58 (51.8%)
Left	54 (48.2%)
Average follow-up period (yr)	6.1

Table 2: Distribution of STA and PAA

Distribution	No. (%)
STA	
A	380 (33.2%)
B1	254 (22.2%)
B2	139 (12.2%)
B3	250 (21.8%)
C1	33 (2.8%)
C2	26 (2.3%)
C3	26 (2.3%)
D1	19 (1.7%)
D2	17 (1.5%)
Total	1144 (100%)
PAA	
1	563 (49.2%)
2	127 (11.1%)
3a	162 (14.2%)
3p	267 (23.3%)
4a	18 (1.6%)
4p	8 (0.6%)
Total	1144 (100%)

sex-orientation follow-up period. Given the lack of follow-up transfemoral cerebral angiography in 3 patients having undergone PAA-EDAS and the failed matching of 1 patient having undergone PAA-EDAS, angiography for a total of 6 patients having undergone PAA-EDAS and 12 having undergone STA-EDAS was reviewed. On the basis of 1-year postoperative medical records, the functional status of the patients was assessed using the following 3 assessments: the clinical outcome grading system and the Karnofsky Performance Scale and mRS scoring methods.[1,17,18] This study protocol was approved by the institutional review board (No. H-1806–116-952).

RESULTS

Table 1 depicts the enrolled patients' demographics; 572 subjects comprised 255 male (44.6%) and 317 female (55.4%) patients. The median age of first diagnosis was 8 years (range, 9 months to 18 years). Of the 572 patients, 112 patients (19.6%) were initially found to have unilateral MMD, with 58 patients (51.8%) being right-sided and 54 patients (48.2%) being left-sided. The average follow-up period was 6.1 years.

As described in Table 2, the most abundant STA class was A (380 hemispheres, 33.2%), 33 hemispheres (2.8%) were class C1, and 19 hemispheres (1.7%) were class D1. For the PAA, the most abundant was class 1 with 563 hemispheres (49.2%), 18 hemispheres (1.6%) were class 4a, and 162 hemispheres (14.2%) were class 3a.

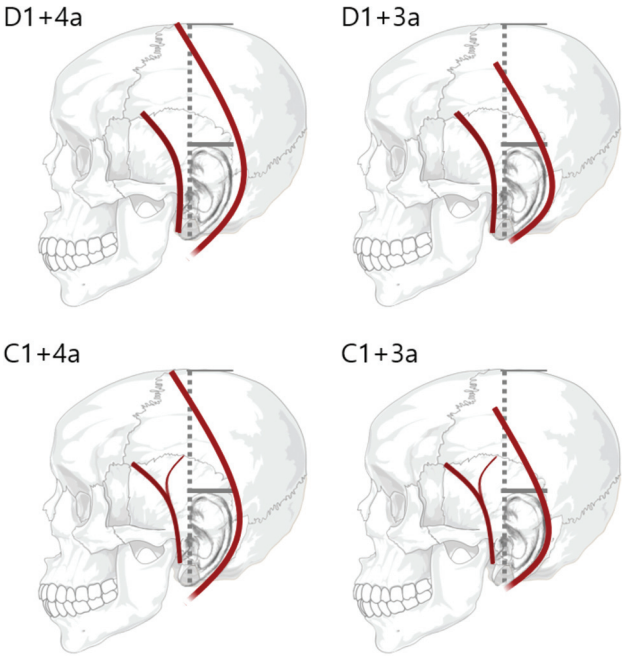

FIG 3. PAA-EDAS candidates.

STA classes A, B, C2, and D2 were considered adequate for performing STA-EDAS. A total of 1066 of 1144 hemispheres (93.2%) had adequate STAs. On the other hand, C1, C3, and D1 were considered inadequate STAs due to the absence or hypoplasia of the parietal branch. A total of 78 hemispheres (6.8%) showed inadequate STAs. Among 78 hemispheres with inadequate STAs, a total of 24 hemispheres derived from 23 patients had STA-PAA combinations of D1 + 4a, D1 + 3a, C1 + 4a, and C1 + 3a, which were considered PAA-EDAS candidates (Fig 3) with 1 patient having bilateral PAA-EDAS candidacy. C3 + 3a and C3 + 4a were omitted as PAA-EDAS candidates because such combinations were not observed in our patients. Among these cases, PAA-EDAS was performed on 10 hemispheres (9 total patients). Three cases with 3a showed PAA terminating over the 50% mark between the helix and vertex (54%, 67%, and 73% for the 3 cases), which was considered adequate for performing PAA-EDAS. The operative cases are listed in the Online Supplemental Data.

For the remaining 14 hemispheres, 4 cases were unilateral MMD, in which the normal side contained STA and PAA configurations of a PAA-EDAS candidacy, and 4 cases were operated as encephalogaleosynangiosis (EGS) due to surgeon's preference. Three cases showed the combination of C1 + 4a in which 1 case had an additional frontal branch that advanced close to the hypoplastic parietal branch, allowing the surgeon to use the frontal branch. For the other 2 cases, a hypoplastic parietal branch was used for STA-EDAS due to surgeon's preference. For the remaining 3 cases that showed a C1 + 3a configuration, the proportion of the PAA termination points between the helix and vertex was below the 50% mark (28%, 17%, and 42% for the 3 cases); hence, the STA parietal branch was used despite being hypoplastic.

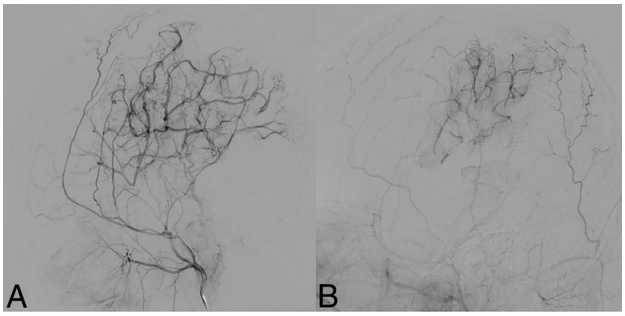

FIG 4. Representative illustrations of late arterial phase external carotid artery angiography with postoperative collateral formation after PAA-EDAS. *A*, Right PAA-EDAS case of 1.7-year-old boy representing Matsushima grade A. *B*, Right PAA-EDAS case of 8-year-old boy representing Matsushima grade B.

For postoperative clinical outcomes, 8 of 9 patients had Karnofsky Performance Scale scores of 100 and mRS = 0, while 1 had a Karnofsky Performance Scale score of 80 with mRS = 2, which was due to an initial preoperative infarction on the left MCA territory. In addition, 6 of 9 patients had excellent clinical outcome, 1 had good clinical outcome, and 2 had fair clinical outcomes.

When we compared the Matsushima grade on follow-up transfemoral cerebral angiography between the PAA-EDAS and STA-EDAS groups, 50% (3 of 6) of the PAA-EDAS and 33% (4 of 12) of the STA-EDAS groups had grade A, 33% (2 of 6) of the PAA-EDAS and 50% (6 of 12) of the STA-EDAS groups had grade B, and 17% (1 of 6) of PAA-EDAS and 17% (2 of 12) of STA-EDAS groups had grade C. Postoperative angiography of illustrative PAA-EDAS cases is presented in Fig 4.

DISCUSSION

Various studies regarding the anatomic variations of STA have been reported in previous literature. One of the earliest investigations was conducted by Marano et al,[13] who classified 10 STA variations based on 50 consecutive human postmortem specimens. Thereafter, Medved et al[14] reported 11 STA variations using a more advanced radiologic technology of digital subtraction angiography on 93 individuals. Regarding ethnic STA variations, Chen et al[19] reported differences between East Asians and whites by comparing the location of STA bifurcation. Kim et al[20] explored STA variations in the Korean adult population using 3D CT angiography, concluding that the STA of Koreans is similar to that of the Chinese, while smaller in diameter compared with that of the Western population. However, a thorough investigation of STA variations in the pediatric population has not been illustrated in the literature. When we compared the results with that of Medved et al,[14] our data also showed that class A (33.2%) was one of the abundant types. However, the proportions of classes B (56.2%), C (7.4%), and D (3.2%) were radically different because Medved et al reported a lower rate of class B (8.6%) and higher proportions of classes C (22.6%) and D (10.8%). These findings may be due to the patients with MMD developing vault Moyamoya collaterals, which could have caused additional formation of frontal and parietal branches.

PAA is well-recognized in plastic and reconstructive surgery for its frequent use in pedicled flaps.[21] However, limited studies have been conducted on its neurosurgical use. Although in 2015, Tokugawa et al[15] coined a novel classification of PAA variations based on the angiographic appearance of 424 subjects, the PAA variation is yet to be discussed for children. On the basis of our study on pediatric patients, classes 3 and 4, the 2 viable candidates for PAA-EDAS, were at 37.5% and 2.2%, respectively. These results were similar to the adult data from Tokugawa et al, which reported classes 3 and 4 to be 48.8% and 1.2%, respectively. However, Germans et al[10] and Pinar et al,[22] respectively, reported 5.7% and 33% of dominant PAAs reaching up to the parietotemporal area in the adult population. This wide discrepancy may be due to ethnic differences, but the dominant PAA described by Germans et al and Pinar et al included classes 3 and 4 combined, which could yield results similar to that of our data when adjusted for the classification suggested by Tokugawa et al. On the basis of the results, we believe that adults and children do not differ in the formation of PAA.

To treat MMD, establishment of sufficient revascularization to the ischemic brain tissue is essential, especially in developing brains. Classically, the STA parietal branch has been used for MCA territory revascularization. Recent studies, however, have reported the potential use of the PAA for MCA territory revascularization in certain cases in which the STA is unfit to be a donor. To use the PAA as a donor, Germans et al[10] mentioned that the PAA must extend to the temporoparietal area with a diameter of at least 1 mm. Similarly, Tokugawa et al reported that an adequate PAA must have a diameter of >1 mm with a length >70 mm from the superior margin of the zygomatic arch. Although both studies briefly mentioned the association of STA and PAA anatomic variations, a close analysis of STA and PAA variants has not been conducted. Hence, by implementing anterior advancement of the PAA through modification of the classification suggested by Tokugawa et al and combining such variants of PAA with certain STAs, we devised 4 combinations of STA and PAA variants that could be considered potential PAA-EDAS candidates. The PAA diameter was not considered in this study because subjects underwent indirect revascularization via EDAS. In addition, C3 (both hypoplastic frontal and parietal STA branches) was omitted as a viable candidate of PAA-EDAS because none of the cases had STA-PAA combination of C3 + 3a or C3 + 4a. This could be because despite the frontal and parietal branches being considerably narrower than the STA trunk, most parietal branches of C3 were suitable to be used as the donor. Unlike the hypoplasia of C1, in which most blood flows toward the dominant frontal branch, making the parietal branch truly hypoplastic, the hypoplastic parietal branch of C3 seemed to deliver a sufficient amount of blood flow, which is supported by the fact that the dominant PAA was not formed in patients with STA class of C3.

A total of 24 hemispheres from 1144 (2.1%) were considered PAA-EDAS candidates. Only 1 patient of 23 subjects had bilateral STA and PAA variants adequate for consideration as a PAA-EDAS candidate. Given the scarcity of intrapersonal bilateral PAA-EDAS candidacy, the intrapersonal development of STA and PAA variants seems mutually independent of each hemisphere. Of the 24 hemispheres, 10 underwent PAA-EDAS (41.7%). We believe that the

proportion of PAA-EDAS could have been increased if adequate combinations of STA and PAA were located in the ipsilateral hemisphere of the patients with unilateral MMD because 4 cases of unilateral MMD had STA-PAA combination patterns on the normal hemisphere. Additionally, 4 of the 24 hemisphere candidates were treated with EGS instead of PAA-EDAS. According to our data base, the last PAA-EDAS candidate to receive EGS was in 2009, and considering the recent use of PAA for extracranial-intracranial bypass in the literature,[9-12] the numbers for PAA-EDAS could have been increased had EDAS been performed on the 4 cases that underwent EGS. Hence, although PAA-EDAS candidacy may be rare, the STA and PAA variant combinations we devised seemed to sufficiently reflect the opportunity for PAA use in performing EDAS, giving surgeons more options for appropriate treatment.

The efficacy of PAA-EDAS compared with STA-EDAS has not been studied in the previous literature. Hence, we compared the Matsushima grade of 6 patients having undergone PAA-EDAS with that of the 12 subjects having undergone STA-EDAS matched for the age-sex-orientation follow-up period. In both PAA-EDAS and STA-EDAS groups, 83% of subjects had grade A or B on follow-up transfemoral cerebral angiography. In the PAA-EDAS group, based on the 1-year postoperative records, 6 patients were asymptomatic, 1 patient had a fixed neurologic deficit but was asymptomatic in terms of ischemic symptoms, and 2 patients postoperatively showed subsided intensity and frequency of ischemic symptoms, such as TIA and headaches. Also, no patient having undergone PAA-EDAS showed deterioration on the Karnofsky Performance Scale and mRS. Thus, the postoperative clinical course of PAA-EDAS does not appear inferior to that of STA-EDAS.

There are a few limitations to this study. First, due to the wide spectrum of class 3 PAA variants, some PAAs failed to adequately reach the parietotemporal area to be used as an STA alternative, despite subjects having D1 + 3a and C1 + 3a STA-PAA variants. Hence, we analyzed the proportion of PAA termination points between the helix and vertex for cases that used the subclassification 3a and cases that did not. Review of the data for D1 + 3a and C1 + 3a groups showed that in all cases using subclassification 3a as the donor, the PAA terminated above the 50% mark between the helix and vertex. On the contrary, in cases that used the hypoplastic STA parietal branch as the donor, subclassification 3a terminated below the 50% mark. On the basis of this result, we believe that further subclassification of class 3a based on its adequacy may be helpful in surgical planning.

Second, due to the scarcity of adequate PAAs, the number of subjects having undergone PAA-EDAS and corresponding STA-EDAS subjects was limited. Consequently, the clinical outcome of patients having undergone PAA-EDAS may have been prone to selection bias.

Nevertheless, this study with the largest number of patients enrolled is the first to comprehensively investigate the anatomic variations of both STA and PAA in pediatric patients with MMD. Also, close observation of the relationship between STA and PAA variants has been made to discern the potential candidates for PAA-EDAS. Finally, in comparison of the PAA-EDAS and STA-EDAS clinical outcomes, the prognosis of PAA-EDAS is not inferior to that of STA-EDAS.

CONCLUSIONS

On the basis of the results, with the appropriate combination of STA and PAA variants, we strongly recommend that surgeons consider MCA territory revascularization with the use of the PAA when the STA parietal branch is unfit for use.

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