ISSN: 2754-4737

Journal of Neurology Research Reviews & Reports

SCIENTIFIC Research and Community

Case Report

Open Access

Encephaloduroarteriosynangiosis (EDAS) in a 31-Year-Old Filipino Female with Moyamoya Disease: A Case Report

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ABSTRACT

Introduction: Adult Moyamoya Disease is a rare chronic cerebrovascular disorder presenting with narrowed or stenotic vasculatures presenting as hemorrhagic stroke in adults. In the Philippines, stroke is a leading cause of death and disability in adult population.

Objectives: This case report aims to discuss the course and clinical outcome of an adult with Moyamoya Disease who underwent Encephaloduroarteriosynangiosis (EDAS) for indirect revascularization and explore decision points for such surgical management.

Case: The index patient is a 31-year-old female who presented with headache, visual disturbances, right-sided weakness, anterograde amnesia, retrograde amnesia, and cognitive impairment. Patient had severe cognitive impairment, no light perception on both eyes, with right hemiparesis of 4/5 motor strength. Preoperatively, patient was prepared for a double set-up direct and/or indirect revascularization technique (STA-MCA Bypass surgery and/or EDAS) however intraoperatively there was a note of 0.6-0.7mm diameter of the left parietal branch of the Superficial Temporal Artery which led to the decision to perform indirect revascularization i.e., Left Encephaloduroarteriosynangiosis (EDAS).

Discussion: Adult cases are ideally managed using a combined approach of direct and indirect revascularization techniques. But for this case, the donor vessel was not suitable for STA-MCA bypass surgery hence the performance of EDAS. Collaterals from EDAS were formed with the left side middle cerebral artery territory.

Conclusion: Multidisciplinary Approach plays a significant impact involving the Cerebrovascular Neurosurgeon, Endovascular Neurosurgeon, Neurologist, Neuroradiologist, Ophthalmologist, and Physiatrist in this field to define optimal screening and treatment strategies in patients with Moyamoya Disease. Six months after surgery, patient had improvements in the cognitive, neurologic status, radiographic, and functional outcome with modified Rankin Scale Score of 1. Patient remained stroke free since the time of the operation. Hence, indirect revascularization technique (EDAS) appears to be a safe and promising intervention even for adult patients with Moyamoya Disease.

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Received: July 27, 2026; **Accepted:** August 06, 2026; **Published:** August 17, 2026

Keywords: Encephaloduroarteriosynangiosis, Moyamoya Disease, Revascularization, STAp (Superficial Temporal Artery Parietal Branch)

Introduction

Adult Moyamoya Disease is a rare cerebrovascular disorder presenting with narrowed or stenotic vasculatures of the internal carotid, anterior and middle cerebral arteries [1]. The natural history of the disease varies per patient. Usually, in adult population, Moyamoya disease presents as hemorrhagic stroke in contrast with the pediatric age group presenting as ischemic stroke [2]. In the Philippines, stroke is the second leading cause of death and the first leading cause of disability in adult population [3,4]. The prevalence of Moyamoya Disease is higher in Asian population, noted incidence as 0.35 per 100, 000 population in Japan with case prevalence of 3.16 per 100,000 population and

lower incidence in non-Asian population [5,6].

Surgery is the mainstay of treatment for Moyamoya Disease. Surgical revascularization techniques including direct, indirect, and combined strategies are employed to augment cerebral blood flow and improve cerebral hemodynamic instability [6]. With these, rates of postoperative transient ischemic attacks were low, and no patients had new ischemic or hemorrhagic stroke on follow up [6]. Indirect revascularization is a common and effective treatment for pediatrics but not usually in adult Moyamoya cases [5,7]. Direct revascularization technique alone or in combination with an indirect procedure is considered to be the therapy of choice in adults, as an indirect method alone has been reported to be unpredictable or ineffective to obtain good revascularization5. Several indirect techniques such as Encephalomyosynangiosis (EMS) and Encephaloduroarteriosynangiosis (EDAS) are classifiable according to the tissue (muscle, periosteum, galea, dura mater, and extracranial tissues) or vessel used as a source of blood supply [8]. Postoperatively, follow-up imaging and assessment are done and correlated with good functional and radiographic outcome [9].

The purpose of this case report is to discuss the course of an adult with Moyamoya Disease who was managed with Encephaloduroarteriosynangiosis (EDAS) for indirect revascularization as it explores difficult decision points leading to this surgical management. Thus, specifically aims to:

1. Present a case of a 31-year-old female with Moyamoya Disease.
2. Discuss the intraoperative findings and perioperative course.
3. Illustrate the postoperative radiographic and functional outcome of the patient 6 months after Encephaloduroarteriosynangiosis (EDAS).

Case Report

This is a case of a 31 years old, female, from Pampanga, right-handed, who presented with eleven months history of severe frontal to generalized headache associated with blurring of vision. Patient sought consult and was prescribed with Ibuprofen which afforded temporary relief. Nine months prior to admission, noted recurrence of headache associated with right sided weakness, retrograde, and anterograde amnesia. Patient was admitted at a local hospital and Plain Cranial CT scan showed acute to subacute infarction in the left parieto-occipito-temporal lobes (Figure 1). Patient was managed as ischemic stroke, discharged and improved with right sided residuals, retrograde, and anterograde amnesia. Patient had physical therapy three times in a week for a month which showed improvement, remained well and asymptomatic.

Four months prior to admission, patient experienced recurrence of severe headache associated with right sided weakness and blurring to complete visual loss thus admitted at the local hospital and Cranial MRI with MRA showed encephalomalacic changes from old infarction, left occipital lobe; right temporo-occipital cortical laminar necrosis (Figure 1). Managed as Re-stroke and referred to Rizal Medical Center (RMC) for further evaluation and management.

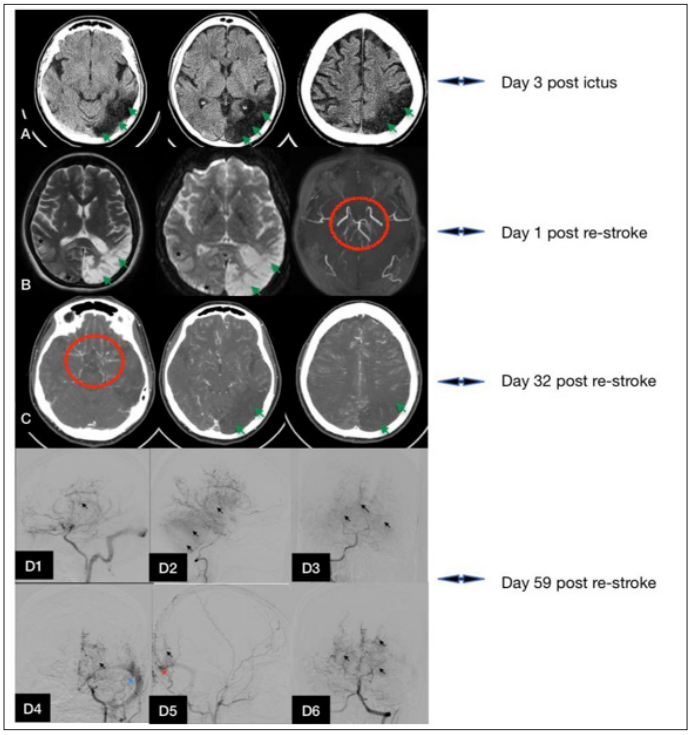


Figure 1: Plain Cranial CT Scan day 3 post ictus (A) showing areas of infarction/hypodensities on the left parieto-occipito-temporal region (green arrows), Cranial MRI with MRA day 1 post re-stroke (B) showing left temporo-occipital old infarction/ encephalomalacic changes (green arrows) and right temporo-occipital cortical laminar necrosis (asterisks), stenotic vessels (encircled red); Cranial CT Angiogram day 32 post re-stroke (C) showing old infarction at the left temporo-occipital region (green arrows) with Moyamoya vessels (encircled red). Digital Subtraction Angiography Day 59 post re-stroke (D) showing Puff-of-smoke or Moyamoya vessels (black arrows). Dural Arteriovenous Fistula of Right Occipital Artery and Torcula (red arrow). Dural Arteriovenous Fistula of Left Temporal Artery and Left Sigmoid Sinus (blue arrow). Left ICA Injection (D1, D4), Right ICA Injection (D2), Right Vertebral Artery Injection (D3), Right ECA Injection (D5), and Left Vertebral Artery Injection (D6).

At RMC, patient had Cranial CT Angiogram (Figure 1) and Endovascular, Transradial, Catheter-based Cerebral Angiography (Figure 1) which showed Moyamoya Disease Stage IV thus advised for STA-MCA Bypass. Digital Subtraction Angiography (Figure 1) revealed severe stenosis with Moyamoya vasculatures on the left internal carotid artery injection. Right internal carotid artery injection showed loss of the normal vessel curvature with Moyamoya vasculatures. Right vertebral artery injection showed the presence of Moyamoya vessels and collateral supply of medial choroidal vessels to the contralateral left parietal cortical region similar findings with the left vertebral artery injection. Right external carotid artery injection revealed collateral supply to left medial frontal lobe with Type I dural arteriovenous fistula of right occipital artery and Torcula with no cortical venous reflux. Left external carotid artery showed dural arteriovenous fistula from left temporal artery and left sigmoid sinus.

Patient is a known hypertensive maintaining on Amlodipine, no previous surgeries, with unremarkable family history. Former Call Center Agent, non-smoker, non-alcoholic beverage drinker, and denies illicit drug use. On examination, patient had stable vital signs, spontaneous eye opening, oriented to person, follows commands, with impaired immediate, recent, and remote memories, MMSE of 5/30 (Severe Cognitive Impairment). Noted light perception on both eyes and right hemiparesis. Managed as Moyamoya Disease Stage IV, Hypertension Stage 2. Medications were given and ensured neuroprotective measures. Patient was referred to IM Neurology, Ophthalmology, and Rehabilitation Medicine for co-management. Preoperatively, Superficial Temporal Artery diameter was 0.5-0.8mm thus patient was prepared for a double set-up direct and/or indirect revascularization technique (STA-MCA Bypass surgery and/or EDAS) however intraoperatively (Figure 2) there was a note of 0.6-0.7mm diameter of the left parietal branch of the Superficial Temporal Artery which led to the decision to perform indirect revascularization i.e., Left Encephaloduroarteriosynangiosis (EDAS).

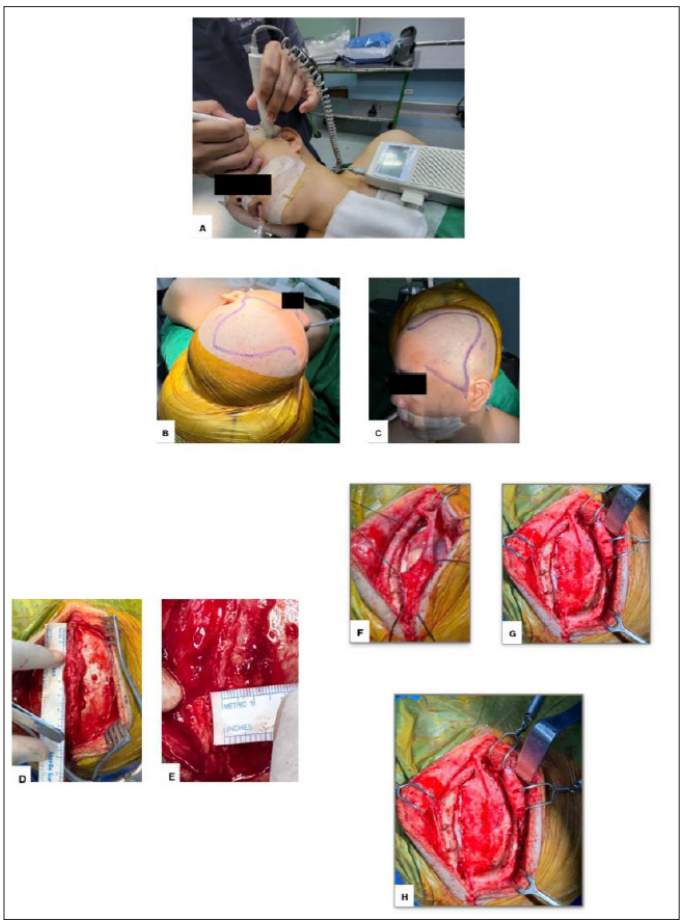


Figure 2: Intraoperative photos of patient during Indirect Revascularization (EDAS). A, Mapping of donor vessel (STAp) using Transcranial Doppler. B, C - planned surgical incision plotted. D, E - donor vessel (STAp) diameter and length evaluated. F, G, H - EDAS completed (vascularized graft with donor anastomosed with dural inversion).

Patient's course in the ward was unremarkable and discharged on the 3rd postoperative day. About 2 months postopertive, patient had improving neurologic status with better motor strength on all extremities. After 6 months postoperative, patient had no complaints, modified Rankin Scale score 1, with improvement in MMSE to 14/30 (moderate cognitive impairment) and significant improvement on the visual acuity from light perception to 20/70 on both eyes.

Patient had Transfemoral, Catheter-based Cerebral Angiography showing new collaterals in the inflow from the Encephaloduroarteriosynangiosis (Figure 3). EDAS formed collaterals with the left side middle cerebral artery territory. There is no progression of Moyamoya vessels. Patient is scheduled for Right Encephaloduroarteriosynangiosis. Informed Consent was obtained from the patient's father and siblings for this report.

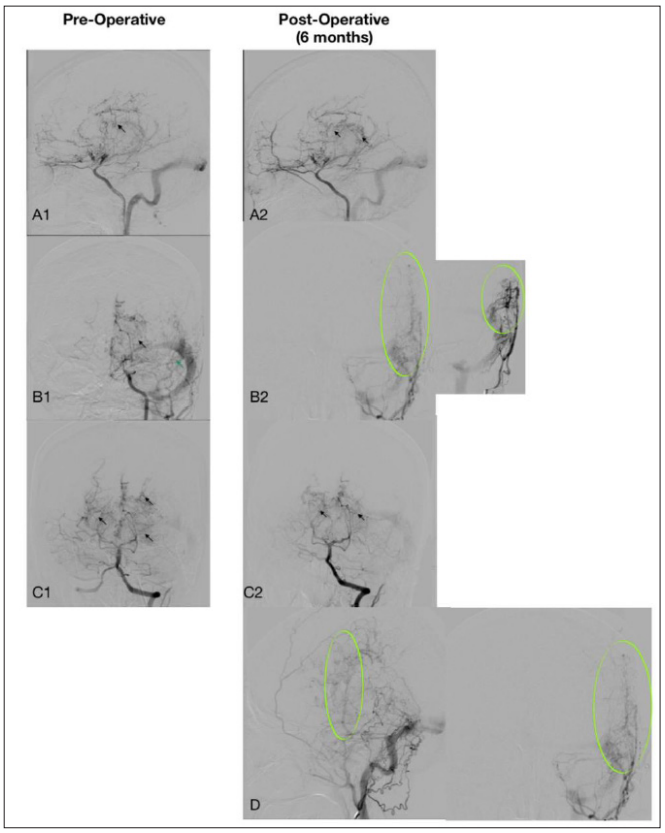


Figure 3: Comparison of Catheter-based Cerebral Angiography Preoperative and 6 months post-operative. Left ICA Injection (A1, A2), Left Vertebral Artery Injection (C1, C2) showing Puff-of-smoke or Moyamoya vessels (black arrows). Left External Carotid Artery Injection (B1, B2, D) showing collaterals from the EDAS (encircled green).

Discussion

The clinical presentation of patients with Moyamoya disease varies considerably. Intracranial bleeding is the typical presentation in adults [5]. The index case in this report manifested as ischemic stroke with no intracranial bleeding noted. Sudden decrease in cognitive performance has been the only clinical presentation in 10% of patients with Moyamoya Disease while the others presented with visual disturbances, blindness, or hemi-convulsive seizures [5,10-12]. The index case presented with headache, visual disturbances, right sided weakness, amnesia, and cognitive impairment suggesting higher cortical involvement. Cerebral angiography is commonly used neuroimaging for Moyamoya Disease which has a crucial role in the six-stage classification of Suzuki and Takaku [10].

Surgical revascularization procedures are performed in Moyamoya Disease to improve cerebral blood flow and hemodynamic status. Surgical options include Direct Revascularization such as STA-MCA

bypass technique, STA-ACA bypass and STA/OA-PCA bypass; and Indirect Revascularization such as Encephalomyosynangiosis (EMS), Encephaloduroarteriosynangiosis (EDAS) and other modifications [13-16]. The index case was proposed for a double set-up direct and/or indirect revascularization technique (STA-MCA Bypass surgery and/or EDAS) considering that the Superficial Temporal Artery diameter was 0.5-0.8mm thus however, intraoperatively, there was a note of stenotic donor vessel with 0.6-0.7mm diameter against the recommended STA diameter of 1mm to 2mm. This led to the decision to do indirect bypass techniques such as Encephalomyosynangiosis which involves implanting the temporalis muscle to the dural edges on the lateral surface of the brain and Encephaloduroarteriosynangiosis which was done and is now the preferred technique [5,17]. In performing EDAS on this patient, there was an exposure and dissection of the parietal branch of the Superficial Temporal Artery (STAp) with preservation of the vascular flow. The donor artery was isolated with surrounding fascia and temporalis muscle was transected to expose the calvarium. Temporal craniotomy was done and the vascularized graft with donor meticulously anastomosed with the dural edges.

Adult cases are ideally managed using a combined approach of direct and indirect revascularization techniques [5]. But for this case, the donor vessel was not suitable for STA-MCA bypass surgery hence the performance of EDAS. Performing EDAS in elderly patients offers low risk of stroke recurrence and an effective way of preventing neurological deterioration thereby possible improving Moyamoya Disease prognosis [11]. EDAS is an indirect revascularization technique proven to be safe and effective for pediatric and adult moyamoya disease with favorable clinical outcomes [17]. The goal of EDAS is to improve the cerebral blood flow so as to prevent further damage or infarction thereby serving as a viable technique in the prevention of appalling neurologic deficits [18-20]. In this report, patient was monitored closely for the next 6 months with note of significant improvements in the cognitive, neurologic status, functional and radiographic outcome.

Conclusion

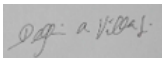
Moyamoya Disease is a challenging disease entity to manage. The constellation of symptoms depends on the structures and vasculatures involved. The clinical presentation may vary accordingly thus Adult Moyamoya Disease can manifest as ischemic stroke even with adult population. Cerebral Angiography is useful in diagnosing Moyamoya Disease. Surgical revascularization technique for Moyamoya Disease should be individualized per patient. Indirect revascularization procedure, although not the therapy of choice in adults, led to a significant good outcome to the index case. Multidisciplinary Approach plays a significant impact involving the Cerebrovascular Neurosurgeon, Endovascular Neurosurgeon, Neurologist, Neuroradiologist, Ophthalmologist and Physiatrist in this field to define optimal screening and treatment strategies. Patient education and strong family support should be a great foundation for further treatment. Recently, patient remained stroke free, independent, cares herself, although with moderate cognitive impairment post-surgery. For adult patients with Moyamoya disease not amenable to direct revascularization, EDAS appears to be safe revascularization procedure with an acceptable 6-month outcome.

Informed Consent

I, the undersigned, hereby authorized Dr. Gimnaste Glenn P Adajar to access my/our patient medical records for his Case Report. Dr Adajar will observe confidentiality along the course of his study and explained the purpose of his work.

As part of our participation for this Research, we will:

1. Comply with the follow up checkup set by Dr. Adajar.
2. Understood that there will be no monetary involvement to both parties.
3. Comply with the whole duration of the study (1 year monitoring)

Guardian/Relative: Delfin A Villa **Signature:** 

Relationship to Patient: Father

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