

CASE REPORT

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Endovascular treatment of distal middle cerebral artery dissecting aneurysm with intracerebral hemorrhage in a paediatric patient: a case report and literature review

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Abstract

Background Intracranial arterial aneurysms in the paediatric age group constitute less than 5% of all aneurysms. The most common form is spontaneous dissecting aneurysms (SDAs) (Peron et al. in *Acta Neurochir* 152(9):1511–1517, 2010; Huang et al. in *Surg Neurol* 63(5):424–432, 2005; Krishna et al. in *Acta Neurochir* 147(5):469–476, 2005; Ihn and Jung in *J Korean Soc Radiol* 65(5):441–445, 2011). It occurs most commonly in the posterior circulation and rarely in the anterior circulation, including the middle cerebral artery (MCA) (Chuang et al. in *Asian J Surg* 35(1):42–48, 2012; Ozaki et al. in *World Neurosurg* 113:208–211, 2018). MCA dissecting aneurysm even rarely presents with intracerebral haemorrhage (Ete et al. in *J Case Rep* 3(2):451–454, 2013; Ahmad in *Interdiscip Neurosurg* 18:100510, 2019; Bartoš et al. in *Brain Sci* 11(1):29, 2020).

Case description A 12 years old non-diabetic, non-hypertensive female presented with distal right MCA (M2-M3 junction) dissecting aneurysm with right parieto-temporal haemorrhage. The patient was well managed with endovascular coil embolization with a good functional outcome.

Conclusion Endovascular coil embolization had favourable clinical and angiographic outcomes in distal MCA dissecting aneurysms.

Keywords Cerebral aneurysm, Arterial dissection, Endovascular, Intracerebral haemorrhage

Introduction

Intracranial arterial aneurysms in the paediatric age group constitute less than 5% of all aneurysms. The most common form is spontaneous dissecting aneurysms (SDAs), followed by saccular, infectious, and post-traumatic aneurysms. Males are more frequently afflicted in the paediatric population [1–3]. The extracranial portion of the internal carotid and vertebral artery is where

the majority of SDAs develop. But intracranial dissecting aneurysms also occur. The posterior circulation, particularly the vertebral and basilar arteries, is the most prevalent site of intracranial dissections [1, 4]. Rarely do SDA ruptures involve the intracranial anterior circulation. Unlike other aneurysms involving branching vessels, SDAs are more likely to affect the truncal region of the vasculature [1].

Subarachnoid haemorrhage (SAH) is a common complication of intracranial dissecting aneurysms of the posterior circulation, and cerebral ischemia/infarct is more common in those of the anterior circulation including MCA. It is extremely uncommon for anterior circulation dissecting intracranial aneurysms to manifest with intracerebral haemorrhage (ICH) [5, 6].

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The vertebrobasilar system has been the focus of the majority of research on aneurysm dissection due to its prevalence. However, for anterior circulation dissecting aneurysms, case reports have been the only source of information. In this article, we present a case of dissecting aneurysm of anterior circulation (M2-M3 junction of the middle cerebral artery) presenting with ICH in a female paediatric patient.

Case description

Non-diabetic and non-hypertensive 12-years-old female presented in the emergency with a history of headache which was sudden in onset and severe in intensity, followed by the loss of consciousness for the last three days. She had weakness in the left half of the body and vomiting for the last two days. The patient had no history of seizures or any systemic infections. There was no history of substance abuse or any history favouring coagulation disorders. No similar history was present in any of the family members. Normal developmental milestones were met by the patient. On presentation Glasgow Coma Scale was E1V1M4, Pulse was 86/min and blood pressure was 112/72 mmHg. There was no neck rigidity. The tone of the left upper and lower limbs was diminished. On the left half of the body, deep tendon reflexes were reduced. The plantar was extensor on the left side. Cranial nerve examination was not possible. Cardiovascular and pulmonary system examinations were within normal limits. In view of the poor GCS and the clinical scenario, the patient was intubated immediately and put on ventilatory support and shifted to the neurosurgery ICU for further evaluation.

A non-contrast CT scan of the brain revealed a large acute intracerebral hematoma in the right parieto-temporal cortex with intraventricular haemorrhage with mass effect (Fig. 1). Routine blood investigations including haemoglobin, total leucocyte count, platelet count, random blood sugar, lipid profile, liver function and renal function tests were within normal limits. The coagulation profile was reported to be within the normal range. The patient underwent 6 vessels selective Digital Subtraction Angiography (DSA) on a biplane DSA machine emergency basis and the report revealed a right middle cerebral artery dissecting aneurysm at the junction of M2–M3. Angioarchitecture of the aneurysm delineated with 3D DSA reconstruction (Fig. 2). The patient was planned for endovascular intervention. The patient underwent serial coil embolization (3 coils; one 6×4, one 4×2, and one 3×2) of the aneurysm with intact distal and lenticulo-striate circulation on check DSA (Fig. 3). On Post-op day 1, the patient was kept intubated and weakness on the left side was the same as that of pre-op. Vitals were within normal range and the patient was

closely monitored in the neurosurgical ICU. Antiedema measures were taken and tablet nimodipine was given through Ryle's tube. Compression stockings were applied for deep venous thrombosis prophylaxis and limb physiotherapy continued. The patient was extubated on post-operative day 4. Serial check non-contrast CT revealed resolving hematoma with gradual improvement in the power of the left upper and lower limbs. On postoperative day 14, the patient's power in the left upper limb was 4/5 and the lower limb was 4+ /5 (Fig. 4).

Discussion

Trauma, drug abuse, coagulation disorders, and cerebral AVM are all common causes of ICH in a non-hypertensive young patient [7]. With the advancement of diagnostic imaging techniques, spontaneous intracranial arterial dissection (IAD) has assumed a greater significance as a cause of strokes. Spontaneous IAD is more common in the posterior circulation and rare in the anterior circulation, which includes the middle cerebral artery (MCA) [8]. The majority of extracranial carotid and vertebral artery dissections have a favourable prognosis. However, intracranial artery dissections are often accompanied by severe neurological impairment and thus have a poor prognosis. Patients who present with haemorrhage may need immediate surgical intervention due to the high incidence of early re-haemorrhage [9]. The goal of the treatment plan should be to prevent the IAD from re-rupture while protecting key perforators close to the lesion and blood flow distal to the dissection [5, 10].

Several surgical and endovascular treatment methods have been documented. But no consensus has been achieved on the best surgical and endovascular therapy options for this condition. Treatment options for dissecting MCA aneurysms include microsurgical clipping and endovascular procedures. The choice of procedure depends on several factors such as the location of dissection, presence of local perforators and collateral supply [11]. Microsurgical clipping is preferred for saccular aneurysms. Microsurgical clipping for dissecting aneurysms is associated with high morbidity [5, 10]. Moreover, nowadays endovascular procedures are preferred over microsurgical approaches because of better clinical outcomes [12].

Options available for endovascular procedures are parent artery occlusion, stent-assisted coiling and conventional coiling. But, the treatment principles of anterior circulation dissecting aneurysm differ from the posterior circulation dissecting aneurysm due to different angioarchitecture and collateral supply. The vertebrobasilar dissecting aneurysm can be treated by occlusion of dissecting aneurysm and parent artery (deconstruction) if the flow from the contralateral vertebral artery is patent

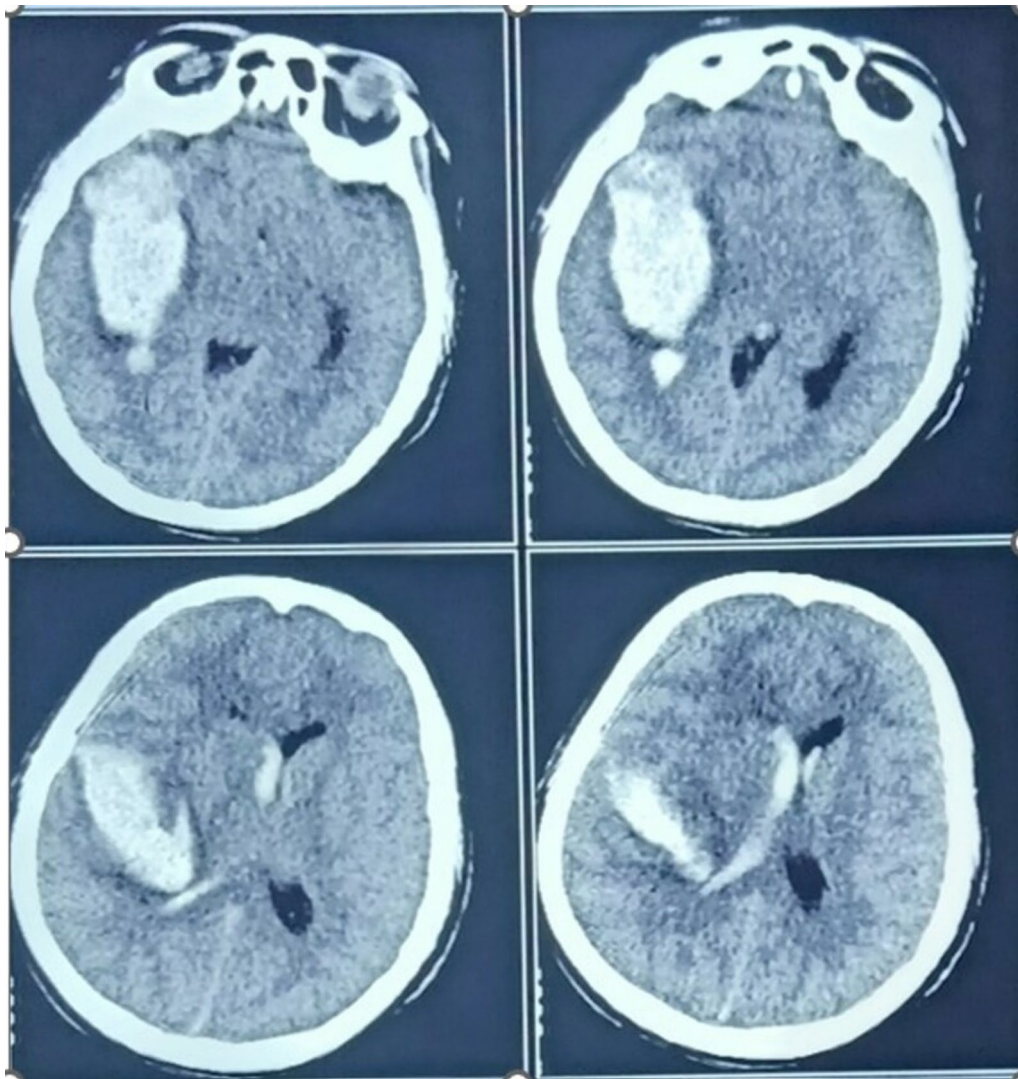


Fig. 1 Non-contrast CT head showing a large acute intracerebral hematoma in the right parieto-temporal cortex with

[6]. Whereas, parent artery occlusion in dissecting an MCA aneurysm has to be accompanied by a bypass using STA-MCA (superficial temporal artery to middle cerebral artery) or RA-MCA (radial artery to middle cerebral artery) to avoid severe MCA territory infarct. This bypass surgery couldn't be done in the pediatric age group due to fragile vessels. Moreover, the clinical outcome depends on the graft patency [13, 14].

For the past few years, flow diverters or intracranial stents have been used as a parent artery reconstruction for dissecting aneurysms. But it has to be carefully considered in pediatric patients because implantation of these devices needs dual anti-platelet therapy throughout the life which is a risk factor for re-haemorrhage. The long-term risks of antiplatelet agents in

the pediatric population are unknown. Moreover, the child's development and the anticipated alteration in the size of the parenchymal vessels might be considered as a factor in preferentially avoiding using these devices [11, 15]. Therefore, few countries don't prefer using these devices in paediatrics. Although, few authors have stated the successful outcome of using flow diverters or stents in paediatrics [16]. Taking note of all these facts and considering the small lumen of distal MCA in our case we refrained from using flow diversion. Several reports have been published for conservative treatment of IADs. But recent studies have suggested that patients with MCA dissecting aneurysms who presented with ICH had better functional outcomes if they underwent surgical intervention than



Fig. 2 a DSA revealed a dissecting distal right MCA aneurysm at the M2–M3 junction, b 3D reconstruction image showing an aneurysm of size 8.03 × 2.84 mm

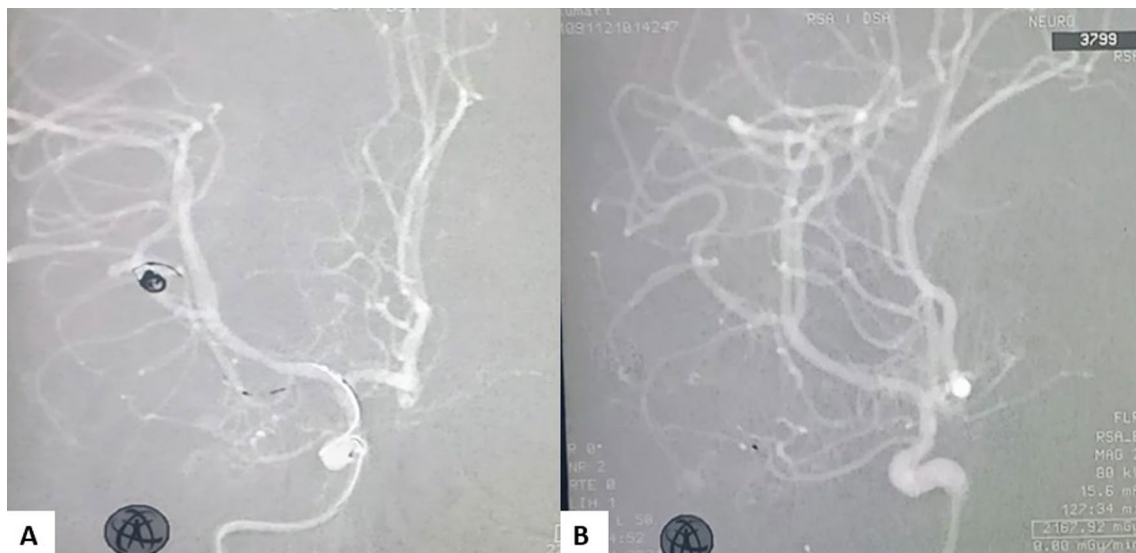


Fig. 3 a Serial coil embolization of the aneurysm, b Post-coiling DSA shows complete obliteration of the aneurysm with intact distal and lenticulo-striate circulation

that conservative treatment [5]. Keeping in mind, the re-haemorrhage risk in conservative strategy, we continued with the conventional coiling of dissecting aneurysm. In this case, we have successfully treated dissecting aneurysms of anterior circulation in the

paediatric age group with atypical presentation and had a favourable clinical outcome.

Demographic profile, clinical data and various modes of intervention in the young age group with dissecting MCA are summarized in Table 1. To the best of my knowledge, atypical presentation of dissecting MCA

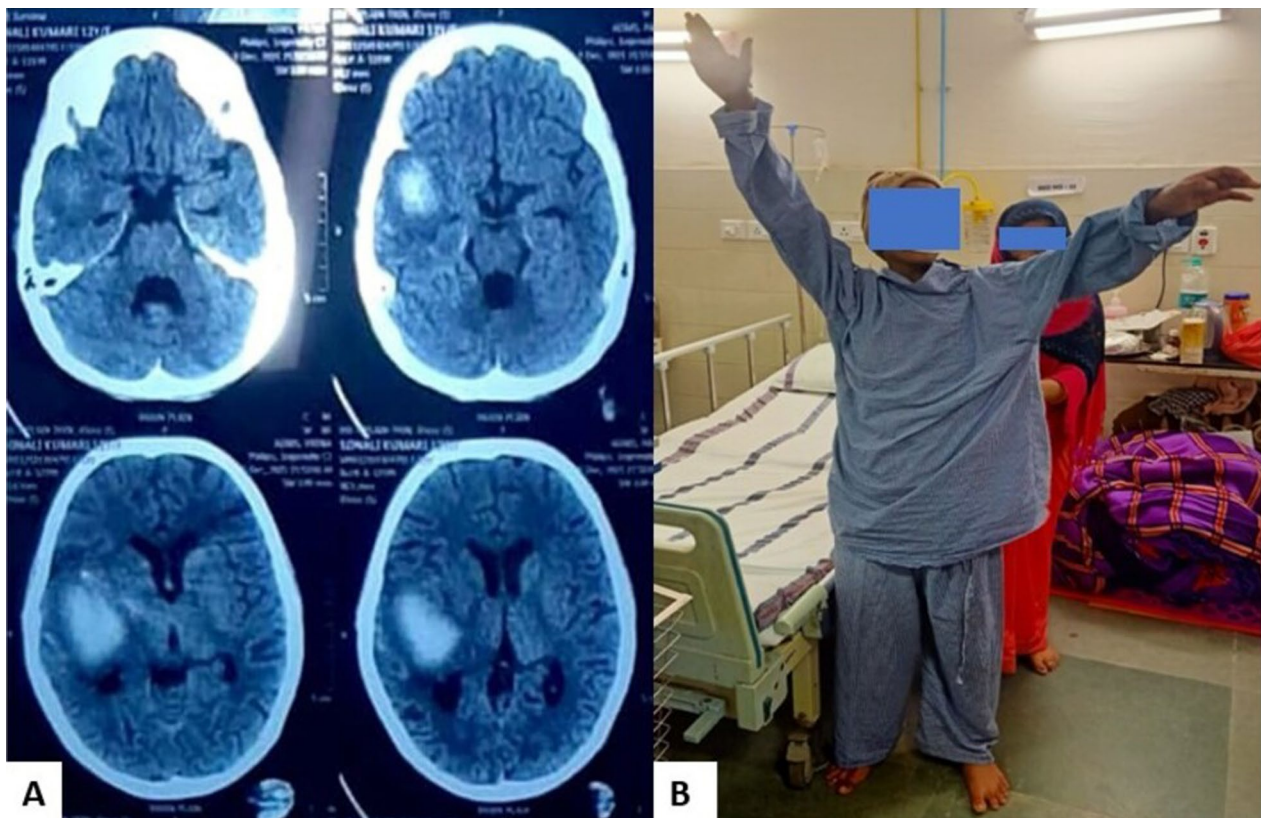


Fig. 4 **a** Non-contrast CT head done on post-op day 8, showed resolving hematoma, **b** Post-op day 14 with clinical improvement in the power of left upper and

Table 1 Summary of illustrative cases of dissecting aneurysm in paediatric age group and comparison with the present case

Study	Cases	Age 9 (yrs.)/sex	Presentation	Location	Procedure	Result
Puyuan zhao et al. [17]	1	22/M	SAH	MCA bifurcation	Parent vessel occlusion	Occluded
Peron et al. [18]	1	1.5/M	ICH	Right M1	Craniectomy with trapping followed by aneurysmectomy	Improved with residual left hemiparesis
	2	8/M	ICH	Left M1	Craniotomy with trapping	Improved with residual right hemiparesis
	3	20/M	SAH	Left M1	Craniectomy with trapping	Improved, no deficit
Tony Ete et al. [19]	1	17/M	SAH	Right M2	Craniectomy with trapping	Improved with Rt. MCA infarct
Zhang et al. [20]	1	18/M	Asymptomatic	Right M1	Conservative	Stable
Fujimura et al. [21]	1	20/M	Intramural hemorrhage	Left M1	Clipping	Improved
Bhagal et al. [11]	1	14/F	SAH with ICH	Left M1	Flow diverter stents	Improved
Present case	1	12/F	ICH with IVH	Right M2–M3	Endovascular coil embolization	Improved

SAH Subarachnoid Hemorrhage, ICH Intracerebral Hemorrhage, IVH Intraventricular Hemorrhage, MCA Middle Cerebral Artery

aneurysm with involvement of distal segment, the feasibility of the endovascular technique and good clinical outcome in such case had been first time described and thus need to be mentioned.

Conclusions

An intracranial artery dissection should be considered as a differential diagnosis in the case of ICH in a young patient who does not have any other known causes, and cerebral angiography can provide valuable information. For MCA dissecting aneurysm, endovascular therapy had favourable clinical and angiographic outcomes.

Abbreviations

SDA	Spontaneous dissecting aneurysms
MCA	Middle cerebral artery
SAH	Subarachnoid hemorrhage
ICH	Intracerebral hemorrhage
DSA	Digital subtraction angiography
3D reconstruction	3-Dimensional reconstruction
AVM	Arteriovenous malformation
IAD	Intracranial arterial dissection
STA-MCA bypass	Superficial temporal artery to middle cerebral artery bypass
RA-MCA bypass	Radial artery to middle cerebral artery bypass
CT	Computerised tomography

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Author contributions

Dr. MSA and Dr. VCJ performed literature research and analysis of the originality of the case. The writing phase was done by the whole team. Dr. VSS was supervised. All authors read and approved the final manuscript.

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Declarations

Ethics approval and consent to participate

Not applicable.

Consent for publication

Informed consent was obtained from the patient included in this study.

Competing interests

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