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Case Report

A child who presented with cerebral infarction: Clipping combined with bypass surgery of a thrombosed giant aneurysm

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ABSTRACT

Background: Cerebral aneurysms are not common among children and most of them are presented with subarachnoid hemorrhage or mass effect. Here, we describe a rare case of a pediatric giant aneurysm presented with cerebral infarction.

Case Description: A 38-month-old boy visited the emergency room due to left hemiparesis and left central type facial palsy. Initial magnetic resonance imaging showed acute cerebral infarction on the right basal ganglia and coronal radiata. Furthermore, a thrombosed aneurysm with a diameter of 30.57 mm at the frontal branch of the right middle cerebral artery was observed. A right pterional craniotomy with Sylvian dissection was performed. Superior and inferior divisions of the frontal branch originating from the aneurysm were identified. The superior division was cutoff from an aneurysm and clipping saving the inferior division was done. Subsequently, end-to-end anastomosis was done between a parietal branch of the superficial temporal artery and a superior division from the aneurysm. No acute complication from the operation was observed. Motor power of the left upper extremity recovered after rehabilitation, while fine motor impairment remained 6 months after the surgery.

Conclusion: This case illustrates successful treatment of a pediatric giant aneurysm with extremely rare presentation of cerebral infarction, under a meticulous surgical plan and *ad hoc* modification.

Keywords: Bypass surgery, Clipping, Pediatric cerebral infarction, Pediatric giant aneurysm

INTRODUCTION

Pediatric cerebral aneurysms are not common and it comprises about 0.5–4.6% of all aneurysms.[3] Compared to adult cases, male predominance and the higher percentage (12–37%) of giant aneurysms are characteristics of pediatric cerebral aneurysms.[8] Most of young age cerebral aneurysms are presented with headaches associated with subarachnoid hemorrhage or seizure. Cranial nerve deficits due to mass effect are also common.[1] Especially, giant intracranial aneurysms, which show almost 80% 5-year mortality, can be challenging to surgically treat in cases of children as wide dissection is difficult due to not fully developed Sylvian fissures.[7] In addition, the small vessel size of children make it difficult to do bypass

surgery. Here, we describe a rare case of a pediatric giant aneurysm presented with infarction, treated by clipping combined with bypass surgery.

CASE DESCRIPTION

A 38-month-old boy, 96 cm in height and 14 kg in weight, visited the emergency room due to gait disturbance and dysarthria started abruptly 1 day before the visit. Left hemiparesis of motor grade III and left central type facial palsy were observed. Initial brain computed tomography (CT) and magnetic resonance imaging (MRI) showed acute cerebral infarction on the right basal ganglia and coronal radiata [Figure 1a]. Furthermore, a thrombosed aneurysm with a diameter of 30.57 mm at the frontal branch of the right middle cerebral artery (MCA) was observed. Temporal branch is bifurcated early in this case. MRI showed a prominent wall enhancement at the superior aspect and partial thrombus in the aneurysm [Figure 1b]. Digital subtracted angiography confirmed a giant saccular aneurysm at the right MCA and there were two efferent arteries (superior and inferior divisions) from the inferolateral aspect of the aneurysm [Figures 1c and d]. Right side MCA territory was filled through leptomeningeal collaterals in posterior cerebral artery angiogram which means chronic flow delay due to the giant aneurysm.

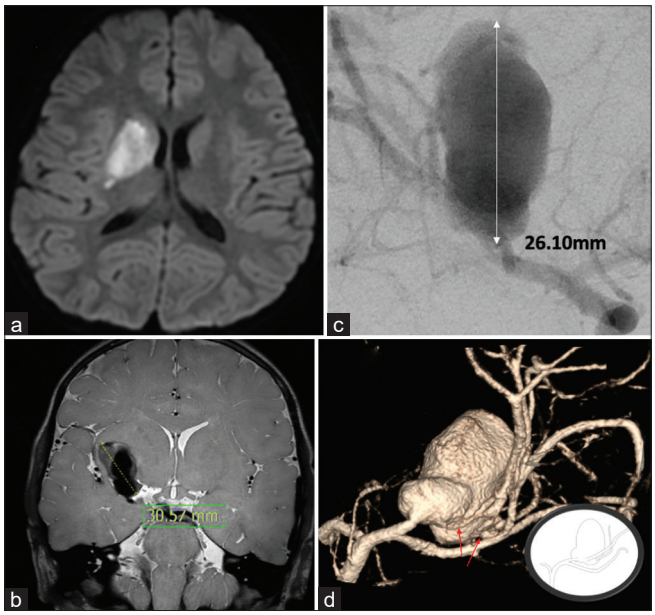

Figure 1: Preoperative images. (a) MR diffusion weighted images at presentation demonstrated cerebral infarction at right basal ganglia and corona radiata. (b) T2 weighted image-vessel wall MR showed aneurysm with partial thrombosis (30.57 mm) and enhancement of superior aspect. (c) Digital subtracted angiography confirmed a large saccular aneurysm (14.9 × 26.1 mm) at the right middle cerebral artery. (d) After early bifurcation of middle cerebral artery, the aneurysm was originated from the frontal branch. Superior and inferior divisions of frontal branch are branching out from the aneurysm dome. (arrows) (inlet).

The patient was delivered at full term, previously healthy, and reached developmental milestones. Echocardiography, blood culture, vasculitis workup, and gene panel were done and these showed no sign of mycotic aneurysm or underlying disease such as coarctation of the aorta, polycystic kidney disease, fibromuscular dysplasia, tuberous sclerosis, Ehlers-Danlos syndrome, and Marfan syndrome. Daily Aspirin of 50 mg (3 mg/kg) was given, and after 1 month, follow-up MRI showed decrease of thrombus in the aneurysm, but size of aneurysm was not changed. Several surgical options were consided based on preoperative images, to obliterate aneurysm and maintain distal flow simultaneously [Figure 2]. The final methed was to be determined according to the findings of the surgery.

After curvilinear skin incision, subgaleal dissection was done to acquire the scalp flap containing the right superior temporal artery (STA). A distal end of STA's parietal branch was cut and prepared for anastomosis. A wide and right pterional craniotomy with Sylvian dissection was performed. Proximal to the aneurysm, we noted prebifurcation of M1. Branches originating from the aneurysm were identified – superior devision of the frontal branch, inferior division of the frontal branch, and lenticulostriate arteries from the frontal branch [Figure 3]. Furthermore, thinned perforators compressed by the aneurysm, a possible cause of infarction, were observed.

Initially, we attempted direct clipping avoiding the superior divison of the frontal branch, but it made the parent artery compromised. Thus, the superior division was cut from an aneurysm and clipping was done for the main sac with one 12 mm fenestrated straight clip. Two 9 mm straight fenestrated clips were used as booster. Inferior division of the frontal branch was preserved [Figure 4]. Subsequently, end-to-end anastomosis was done between a parietal branch of STA and a superior division of the frontal branch from an aneurysm, cut, and prepared in advance [Figure 4]. An

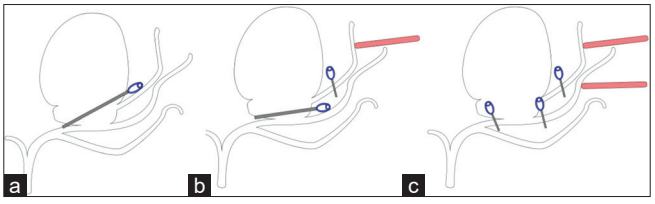

Figure 2: Illustration of operation plan. Gray blade with blue spring represent aneurysm clips. Possible surgical options based on the preoperative images are as follows: (a) direct clipping if possible. (b) Neck clipping combined with anastomosis of superior division of the frontal branch-graft from superficial temporal artery (red bar). (c) Trapping of aneurysm combined with double-barrel bypass via two branches of superficial temporal arteries (red bars).

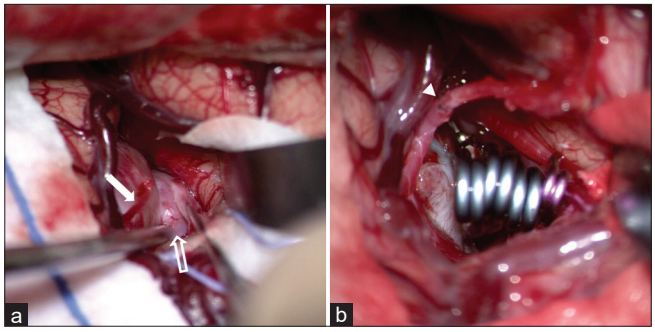

Figure 3: Operative pictures. (a) the aneurysm was originated from the frontal branch of the middle cerebral artery. Superior (solid arrow) and inferior (open arrow) divisions of frontal branch are branching out from the aneurysm. (b) Clipping of main sac was done using multiple fenestrated straight clips. And then the superior division of frontal branch and superior temporal artery's parietal branch were anastomosed in end-to-end fashion. (arrowheads).

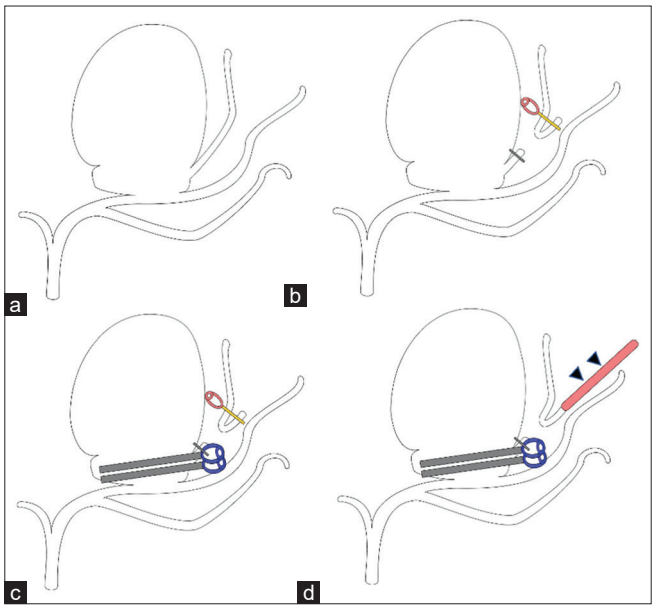

Figure 4: Illustration of operation. Gray blade with blue spring represents fenestrated straight clips while gold blade with violet spring means temporary mini clips. (a) After early bifurcation of middle cerebral artery, the aneurysm was originated from the frontal branch. Superior and inferior divisions of frontal branch are branching out from the aneurysm. (b) Superior division of frontal branch was cut to enable clipping without compromising parent artery. (c) Clipping of main sac was done using multiple fenestrated straight clips. (d) Superior division of frontal branch and distal end of superior temporal artery's parietal branch (arrowheads, red bar) were anastomosed in end-to-end fashion.

additional 3.9 mm mini-J clip was applied to the remnant neck to prevent the inflow around the fenestrated clips. One of lenticulostriate arteries was sacrificed inevitably. After the operation, there was no inflow to the sac and anastomosis was patent under indocyanine green angiography.

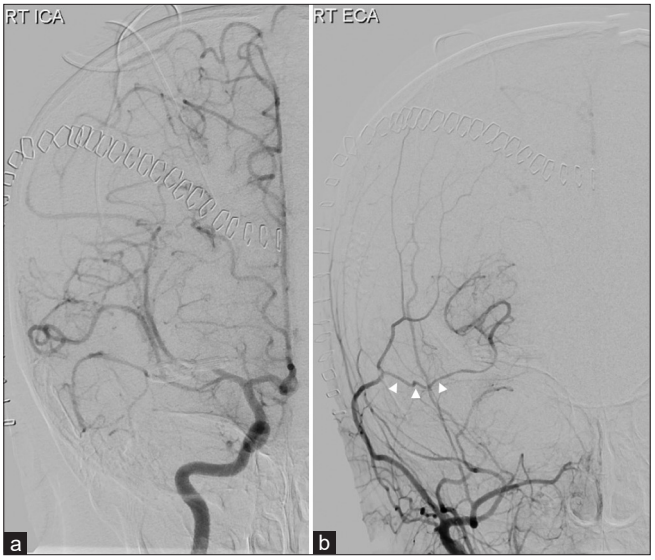

Figure 5: Postoperative images. (a) A digital subtracted angiography at postoperative day 3 showed no residual aneurysmal sac. (b) Patent flow was observed from a graft of superficial artery (arrowheads) to the frontal lobe and insula.

No acute complication from the operation was observed and the patient had no new-onset neurologic deficit. Digital subtracted angiography at postoperative day 3 showed no residual aneurysmal sac and good flow was observed from the donor – right STA parietal branch – to the recipient – M2 area [Figure 5]. After a month, motor power of the left extremity was recovered from grade III to IV after surgery and rehabilitation, and the patient became self-ambulatory. After 6 months, he became active except for fine motor impairment.

DISCUSSION

In this report, we illustrated a rare case of giant pediatric aneurysm that presented as cerebral infarction. One study that reviewed papers on pediatric "giant" aneurysm published from 1990 to 2012 showed that there was no case with cerebral infarction (even though partial thrombosis was present in 23.9% of cases).[8] Furthermore, in Korea, one study based on one tertiary hospital, there were 23 patients with pediatric cerebral aneurysm from 1995 to 2017, but there was no case presented with cerebral infarction.[6] Thrombus from the aneurysm might be the main cause of cerebral infarction in this case. However, squashed and thinned perforator observed during the operation could be the cause. Exceptionally, according to a previous report, about 36% of children who had human immunodeficiency virus-associated cerebral vasculopathy developed cerebral infarction as the aneurysm presentation.[2]

Revascularization techniques involving bypass surgery are one of treatment options for pediatric giant aneurysms, while

clipping is practiced more widely according to the previous reports.[10] Extracranial-intracranial (EC-IC) bypass, which has low complication rate and high patency, can be an adequate choice of bypass technique for complex aneurysms. In children, both vein grafts and STA were reported to be used for EC-IC bypass.[5] Furthermore, revascularization procedure is expected to reduce the risk of late ischemic stroke.[9]

In this case, different surgical approaches were considered preoperatively to achieve the goals of complete ligation of aneurysm and preservation of distal blood flow. Since pediatric aneurysms recur more often than adults counterpart, to minimize the chance of recurrence, complete obliteration was to be achieved. Clipping without bypass was one of the best options [Figure 2a]. However, if this makes remnant at aneurysm neck, clipping combined with bypass to the superior divison of frontal branch was considered as an alternative approach [Figure 2b]. When clipping was not feasible due to atheroma at aneurysm neck, resection of aneurysm after bypass to both superior and inferior division of frontal branch would have been attempted [Figure 2c].

Endovascular treatment was not chosen in this case for a couple of reasons even though the previous studies showed no difference in long-term outcome between endovascular and surgical treatments for ruptured or unruptured pediatric intracranial aneurysms.[11] First, coil embolization for giant thrombosed aneurysm has high recurrence rate and younger age increases the chance for recurrence, given the average life expectancy of 80 years.[4] Second, this patient's infarction could be caused by the mass effect from the compression of perforator. As coil embolization cannot resolve the mass effect, if the aneurysm is giant and thrombosed, surgical approach like bypass might be the better option.[5] Third, the aneurysm had wide neck and a branch out of it, which makes endovascular approach difficult. Therefore, endovascular approach was not a reliable treatment option for our patient.

Sylvian fissure of children is not fully developed and it makes wide dissection difficult. Furthermore, small-vessel size makes bypass surgery challenging. The patient of this case was 38-month-old whose head circumference was 70% of adult head circumference but vessel diameters of recipient candidates were very small (about 1 mm). Moreover, even though the aneurysm was located at the frontal branch of MCA after the bifurcation, as it was bifurcated early, small perforators like lateral lenticulostriate arteries were present. A small perforator was sacrified inevitably, but it did not cause additional neurologic deficit.

CONCLUSION

We illustrated the successfully treated case of a giant MCA aneurysm of a very young age, with an extremely rare presentation – a cerebral infarction. We learned from this case that it is important to make meticulous surgical plan in advance and to modify surgical methods *ad hoc* according to intraoperative situations. This microsurgical solution of bypass surgery combined with clipping might be helpful to other challenging cases of pediatric giant aneurysms.

Declaration of patient consent

Institutional Review Board (IRB) permission obtained for the study.

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Nil.

Conflicts of interest

There are no conflict of interest.

REFERENCES

1. Alamanda VK, Tomycz L, Velez D, Singer RJ. Direct, high-flow bypass for a pediatric giant, fusiform aneurysm of the inferior division of M2: Case report and review of literature. J Surg Tech Case Rep 2012;4:53-7.
2. Baeesa SS, Bakhaidar M, Almekhlafi MA, Madani TA. Human immunodeficiency virus-associated cerebral aneurysmal vasculopathy: A systematic review. World Neurosurgery 2016;87:220-9.
3. Jian BJ, Hetts SW, Lawton MT, Gupta N. Pediatric intracranial aneurysms. Neurosurg Clin N Am 2010;21:491-501.
4. Kalani MY, Elhadi AM, Ramey W, Nakaji P, Albuquerque FC, McDougall CG, *et al*. Revascularization and pediatric aneurysm surgery. J Neurosurg Pediatr 2014;13:641-6.
5. Mura J, Torche E, Riquelme F, Parra M, Julio R. Three-year-old patient with giant MCA aneurysm treated by trapping-resection plus STA-MCA bypass. Case report. Childs Nerv Syst 2012;28:169-73.
6. Nam SM, Jang D, Wang KC, Kim SK, Phi JH, Lee JY, *et al*. Characteristics and treatment outcome of intracranial aneurysms in children and adolescents. J Korean Neurosurg Soc 2019;62:551-60.
7. Parkinson RJ, Eddleman CS, Batjer HH, Bendok BR. Giant intracranial aneurysms: Endovascular challenges. Neurosurgery 2006;59(5 Suppl 3):S103-12; discussion S3-13.
8. Pruvot AS, Curey S, Derrey S, Castel H, Proust F. Giant intracranial aneurysms in the paediatric population: Suggested management and a review of the literature. Neurochirurgie 2016;62:20-4.
9. Schaller B. Extracranial-intracranial bypass to reduce the risk of ischemic stroke in intracranial aneurysms of the anterior cerebral circulation: A systematic review. J Stroke Cerebrovasc Dis 2008;17:287-98.
10. Strickland BA, Attenello F, Russin JJ. Extracranial to intracranial bypass for the treatment of cerebral aneurysms in the pediatric population. J Clin Neurosci 2016;34:6-10.

11. Yasin JT, Wallace AN, Madaelil TP, Osbun JW, Moran CJ, Cross DT, *et al*. Treatment of pediatric intracranial aneurysms: Case series and meta-analysis. J Neurointerv Surg 2019; 11:257-64.

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