SAGE Open Medical Case Reports
Volume 12: 1–6

DOI: 10.1177/2050313X241288427
journals.sagepub.com/home/sco
Sage

A rare case of pediatric distal middle cerebral artery aneurysm

Rachel Moor[1], Chance Fleeting[2], Grace Hey[2], Chandana Karumanchi[2], Jason Blatt[1], Matthew Koch[1] and Jesse Lee Kresak[1]

Abstract

Pediatric middle cerebral artery aneurysms are often complicated by subsequent rupture and subarachnoid hemorrhage. Unruptured middle cerebral artery aneurysms are rare in infancy. Here, we present a rare case of a 13-month-old male who underwent clip ligation for the treatment of an incidental M3 aneurysm.

Keywords

Middle cerebral artery, intracranial aneurysm, pediatrics

Date received: 31 May 2024; accepted: 16 September 2024

Introduction

Ruptured intracranial aneurysms are exceedingly rare in the pediatric population, representing approximately 0.17%–4.6% of all intracranial aneurysms.[1] Rupture, hemorrhage, and significant loss of neurologic functioning are typical manifestations of pediatric intracranial aneurysms, highlighting the importance of timely medical intervention despite their rarity.[2] Several case series have indicated pediatric intracranial aneurysms are more often fusiform and located in the MCA when compared to those in adults.[3,4] Furthermore, pediatric MCA aneurysms tend to incorporate the origins of proximal artery branches and are more likely to develop into giant aneurysms (>25 mm) as compared to adults.[5,6] Thus, the clinical management of MCA aneurysms in pediatric patients is often complex and challenging, requiring a robust, multidisciplinary care-providing team.

Clip ligation is a common therapeutic utilized for the treatment of MCA aneurysms.[7,8] Other management strategies include endovascular treatment, surgical decompression without aneurysm treatment, and medical supportive management only.[9,10] It is, however, important to note that most reported cases of pediatric MCA aneurysms primarily occur in children and adolescents.[11] Reports of MCA aneurysms in infancy are limited to a few case reports, and a majority of MCA aneurysms in infancy are identified in patients presenting with hemorrhage (Table 1).[6,12,13]

Case report

We present a case of a 13-month-old male with an incidental right M3 aneurysm. He initially presented to the emergency department with signs and symptoms of a viral illness. No significant family history relevant to the patient's current symptoms is reported. He was found to have a head circumference greater than the ninetieth percentile, so he underwent a limited sequence HASTE magnetic resonance imaging (MRI) brain. This revealed a round lesion at the right frontoparietal junction, which prompted further investigation. On full sequence MRI brain with and without contrast, this was revealed to be a distal MCA aneurysm (Figure 1(a)). The child underwent a diagnostic angiogram to confirm the diagnosis and aid in surgical decision-making. This demonstrated a 7.2 mm × 5 mm saccular distal M3/4 parietal branch aneurysm (Figure 2(a) and (b)). There were otherwise no abnormalities of the cerebrovasculature noted. Given the superficial and surgically accessible location of the aneurysm, as well as the longevity of clip ligation, the decision was made for the right

[1]Department of Neurosurgery, University of Florida, Gainesville, FL, USA
[2]University of Florida College of Medicine, Gainesville, FL, USA

Corresponding Author:
Rachel Moor, Department of Neurosurgery, University of Florida, Gainesville, FL, USA.
Email: Rachel.moor@neurosurgery.ufl.edu

Table 1. Review of reported distal MCA aneurysm in pediatric patients as of April 2024.

Citation	Symptoms and neurologic signs	Lesion location	Treatment	Outcome
Huang et al.[6]	A 14-year-old male patient presented with headache and partial thrombosis of aneurysm	Left MCA	Clipping and resection with deep hypothermic circulatory bypass	No adverse events reported
Buis et al.[12]	127 pediatric patients with MCA aneurysms were included in this analysis	Left MCA in 52 cases; right MCA in 43 cases	86 patients were treated in this cohort. Treatments included surgical clip application ($n=41$), surgical resection or cauterization of the aneurysm ($n=23$), ligation or trapping of the vessel or the parent vessel ($n=8$), endovascular embolization or coiling ($n=10$), or antibiotics only ($n=4$)	Spontaneous thrombosis ($n=5$) and hydrocephalus ($n=3$) were complications associated with treatment. Treated patients demonstrated 75% survival rates in the first 3 weeks after treatment
Lasjaunias et al.[13]	12 pediatric patients with MCA aneurysms were included in this analysis	Not specified	7 patients with MCA aneurysms were treated in this cohort. Treatments included antibiotics ($n=4$) and clipping ($n=3$)	1 patient in this cohort was lost to follow-up. No adverse events were reported with either therapeutic approaches

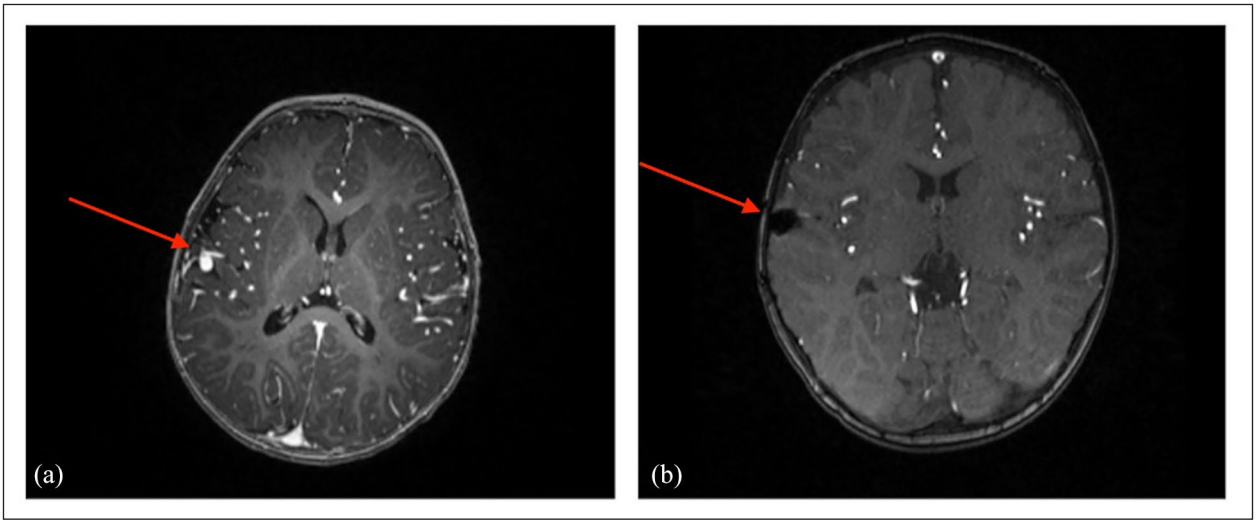

Figure 1. Preoperative MRI with contrast (a) and postoperative MRI angiography (b) demonstrating resolution of the aneurysm (red arrow).

craniotomy for clip ligation. While not urgent, surgery was planned for 1 month thereafter. However, the patient presented preop with a symptomatic respiratory illness, and elective treatment was rescheduled. He ultimately underwent operative intervention 2 months later. The patient underwent a right pterional craniotomy (see attached video). Exposure of the lesion revealed that the proximal vessel appeared plastered to the dome of the aneurysm and plugged into the dome of the aneurysm, with the outflow going to the temporal lobe. There was noted to be an intervening aneurysm dome between the inflow and the outflow of the aneurysm. A clip reconstruction was attempted given this configuration but was unsuccessful given the size and location of the lesion. Reanastomosis was attempted, but vessel length prohibited the success. Ultimately, permanent clips were placed at the aneurysm inflow and outflow, and the dome was carefully dissected and removed. This was sent to pathology (see pathology section for greater detail) (Figure 3). Intraoperative indocyanine green was utilized to demonstrate flow and to allow for manipulation of the clips as desired. Genetic testing was not performed on the aneurysm tissue. Postoperative MRI revealed no stroke or sequela of ischemia. The patient was seen in follow-up several times; he remains neurologically intact. MRI/magnetic resonance angiogram completed 6 months postoperatively without evidence of recurrence or de novo aneurysm formation (Figure 1(b)). Follow-up will be annual moving forward.

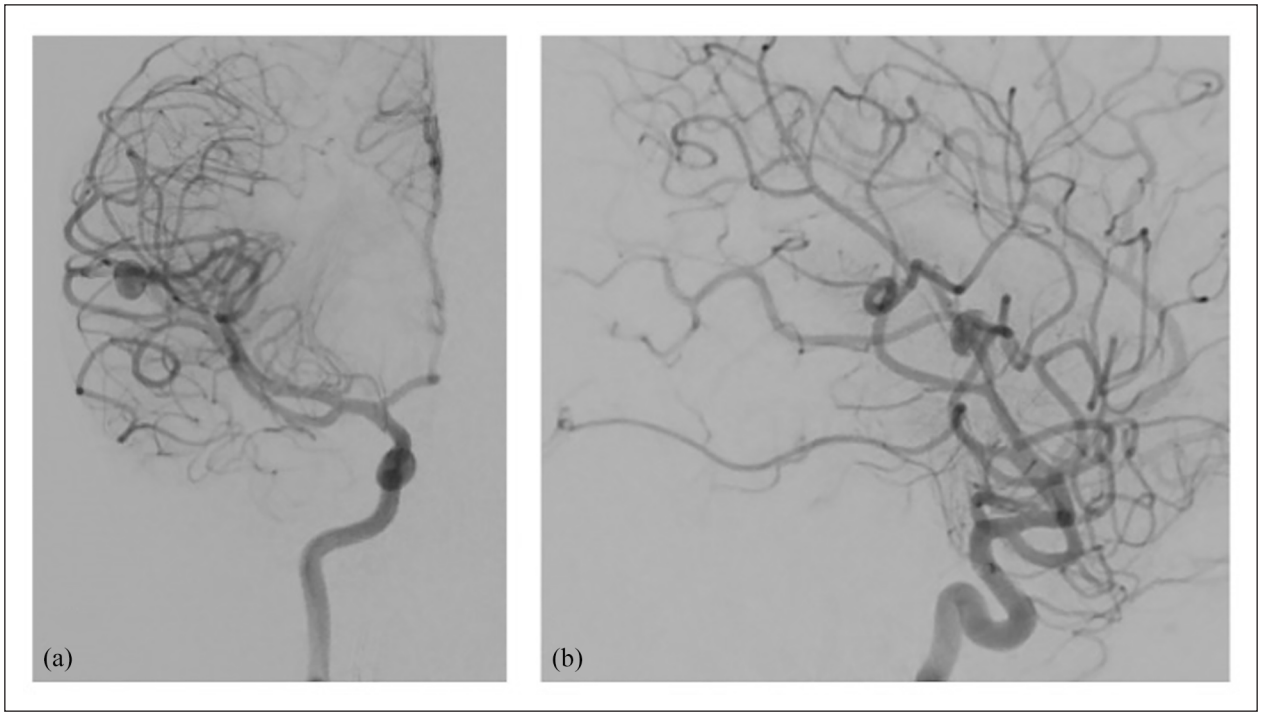

Figure 2. AP (a) and oblique (b) right ICA representative angiographic images demonstrating the distal right MCA aneurysm.

Discussion

Considering the rarity of pediatric MCA aneurysms, it is crucial to discuss the clinical course and management of such cases to expand our understanding of this condition and improve therapy. The optimal management strategy remains unclear; However, within this case, permanent clip ligation is found to be successful without immediate adverse sequelae. This supports the findings reported by Clarke et al.[7] in their analysis of pediatric intracranial aneurysms. Investigating articles spanning 40 years between 1980 and 2020, they found that open surgical aneurysm ligation made up the majority of cases, with a predominance of cases involving idiopathic aneurysms.[6] When compared to adults, clip ligation for MCA or anterior communicating artery aneurysms was found to convey more favorable long-term outcomes in infant patients.[6] Notably, in this case, the lesion was complex in nature, even within the scope of pediatric aneurysms, incorporating fused vessels and compound aneurysmal domes. The success of this treatment thus further implies the generalizability of clip ligation as an effective first-line treatment when such a lesion is detected.

Furthermore, outside of infectious, neoplastic, and traumatic induction, for which this patient has no indication, the cause of pediatric aneurysms remains a controversial topic and is often reported as idiopathic or possibly congenital relating to various discrete defects and genetic conditions, such as polycystic kidney disease, tuberous sclerosis, Ehlers-Danlos, and Marfan syndrome.[14] These unclear origins, coupled with the asymptomatic nature of this form of aneurysm, present the risk that it may go undetected until rupture, posing it as an insidious threat. Specifically, it has been demonstrated that children with ruptured aneurysms experience higher rates of subarachnoid hemorrhage, hematoma, seizures, prolonged hospital stays, and poor outcomes overall when compared to children with unruptured aneurysms.[15–18]

Due to the rarity of these lesions, there is little literature on the histopathologic changes in pediatric or pediatric intracranial aneurysms. Increased inflammatory cells and decreased smooth muscle cells have been associated with an increased risk of rupture in the intracranial aneurysms of adults.[19,20] In the adult aortic setting, there have been investigations into whether certain histologic findings are associated with syndromic aneurysms versus other etiologies, with some evidence to suggest there may be subtle differences.[21] In this report, the absence of key structural components including the tunica intima, tunica media, and elastic lamina suggests the etiology of this patient's aneurysm is likely congenital in nature.[22,23] Further histologic studies may lead to an enhanced understanding of the pathophysiology of pediatric aneurysms and may reveal biomarkers useful for predicting the risk of rupture.

While screening explicit prevention would be ideal, there does not appear to be a single central etiology of pediatric MCA aneurysms that can be leveraged. Short of imaging every child born with a corollary condition, there is currently no explicit mode of detection of unruptured idiopathic aneurysms outside of ancillary and fortuitous findings.[24] That being said, when aneurysms are detected, early treatment, such as that opted for in this case, is vital for the prevention of lasting complications or sequelae up to and including

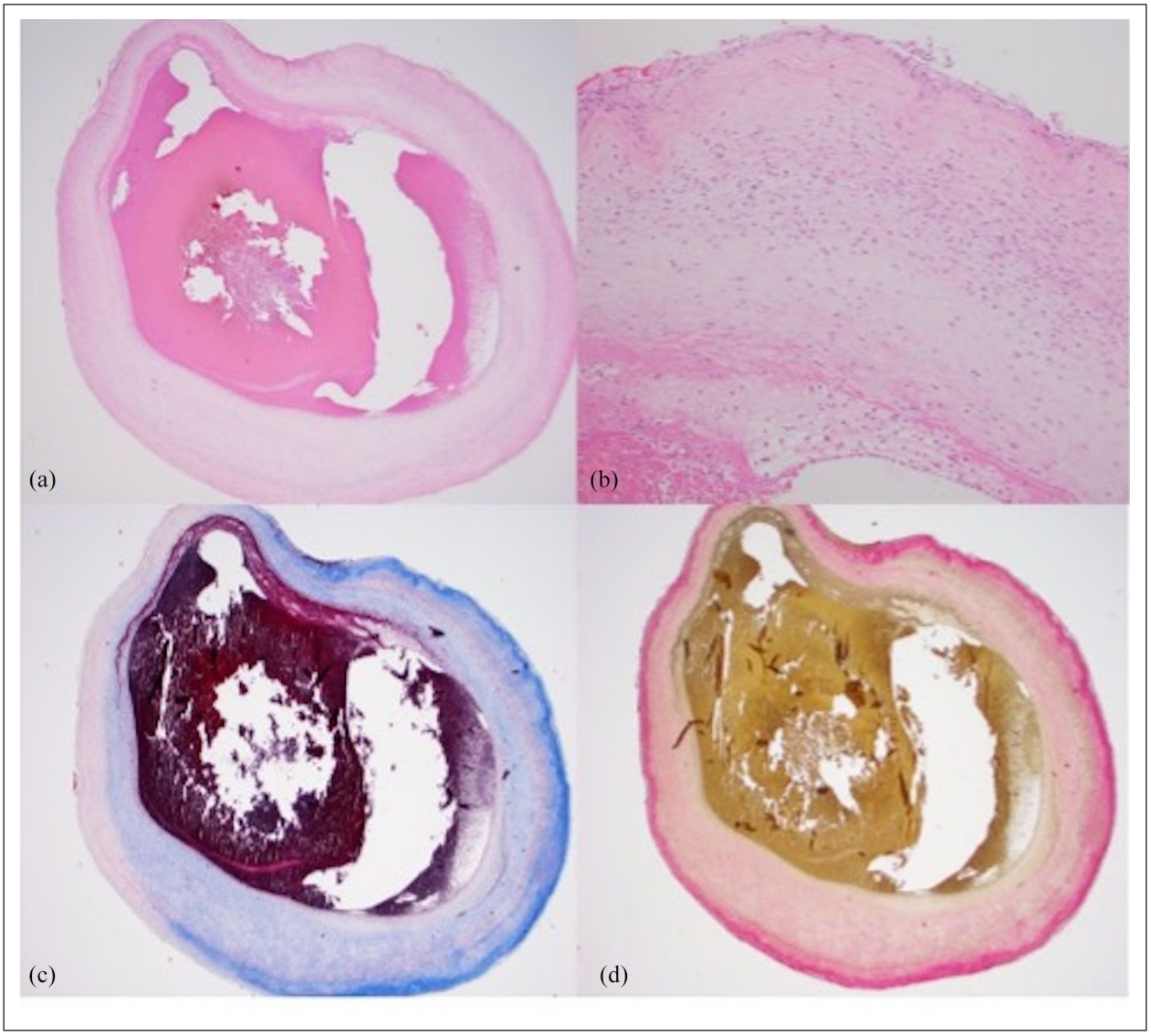

Figure 3. *Pathology*: Hematoxylin-and-eosin-stained sections revealed a segment of the artery with significant alterations of the vessel wall and focal thinning (a). There is little to no tunica intima or tunica media (b). Rather, the wall is predominantly adventitia or fibrous tissue, as highlighted by trichrome stain (c). An absent internal elastic lamina is confirmed by Verhoeff–Van Gieson elastin stain (d).

possible death via rupture/hemorrhage. More research is necessary to understand the underlying pathogenesis and identify risk factors for developing pediatric MCA aneurysms.

In general, pediatric cerebral angiography, endovascular treatment, and open surgery are safe procedures with low rates of major and permanent complications.[25,26] However, it is important to note that these procedures still convey certain risks, particularly for pediatric patients. Non-neurologic complications such as groin hematoma and femoral artery occlusion have been observed following cerebral angiography in children younger than 3 years of age.[27] Complications following endovascular treatment may include vasospasm, though these risks remain relatively unexplored in infancy.[28] Hemiparesis, infection, and subdural fluid collection requiring shunt placement are complications related to craniotomy in infants, though these complications are similar to patients of all ages receiving open surgery for various pathologies requiring neurosurgical intervention.[29–31] Additional research should be conducted to better understand specific risk factors for patients receiving the aforementioned treatments in infancy.

Overall, this case provides an example in which the incidental detection of a rare and unexpected life-threatening condition permitted effective and successful treatment before the onset of any devastating complications. Furthermore, the case presented highlights the inherent complexity and considerations required in managing MCA aneurysms in infancy

and supports the efficacy of clip ligation. Future research and more case reports are necessary to better understand the pathogenesis of unruptured MCA aneurysms in infants to ultimately develop and refine optimal therapeutic strategies.

Conclusion

The prevalence of unruptured distal MCA aneurysms in the pediatric population is exceedingly rare. We have described the unique case of a 13-month-old male treated for an incidental right M3 aneurysm via clip ligation. This report contributed to the limited body of research regarding the presentation of unruptured infantile MCA aneurysms before the onset of devastating neurologic complications.

Acknowledgements

None.

Declaration of conflicting interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.

Ethics approval

Our institution does not require ethical approval for reporting individual cases or case series.

Informed consent

Written informed consent was obtained from a legally authorized representative(s) for anonymized patient information to be published in this article.

ORCID iD

Grace Hey https://orcid.org/0000-0002-9838-7738

References

1. Pollo C, Meagher-Villmure K, Bernath MA, et al. Ruptured cerebral aneurysm in the early stage of life—a congenital origin? *Neuropediatrics* 2004; 35(4): 230–233.
2. Magid-Bernstein J, Girard R, Polster S, et al. Cerebral hemorrhage: pathophysiology, treatment, and future directions. *Circ Res* 2022; 130(8): 1204–1229.
3. Proust F, Toussaint P, Garniéri J, et al. Pediatric cerebral aneurysms. *J Neurosurg* 2001; 94(5): 733–739.
4. Chen R, Zhang S, You C, et al. Pediatric intracranial aneurysms: changes from previous studies. *Childs Nerv Syst* 2018; 34(9): 1697–1704.
5. Fulkerson DH, Voorhies JM, Payner TD, et al. Middle cerebral artery aneurysms in children: case series and review. *J Neurosurg Pediatr* 2011; 8(1): 79–89.
6. Huang J, McGirt MJ, Gailloud P, et al. Intracranial aneurysms in the pediatric population: case series and literature review. *Surg Neurol* 2005; 63(5): 424–432.
7. Clarke JE, Luther E, Oppenhuizen B, et al. Intracranial aneurysms in the infant population: an institutional case series and individual participant data meta-analysis. *J Neurosurg Pediatr* 2022; 30(1): 78–88.
8. Wan KR, Kirollos RW, Lee HY, et al. Giant aneurysm arising from anomalous branch of the middle cerebral artery in a pediatric patient: case report and review of the literature. *World Neurosurg* 2019; 128: 165–168.
9. Burrows AM, Zipfel G and Lanzino G. Treatment of a pediatric recurrent fusiform middle cerebral artery (MCA) aneurysm with a flow diverter. *Case Rep* 2012; 2012: bcr2012010478.
10. Savastano LE, Chaudhary N, Gemmete JJ, et al. Stent-assisted coil embolization of a symptomatic middle cerebral artery aneurysm in an infant. *J Neurosurg Pediatr* 2014; 14(5): 550–554.
11. Kalani MYS, Elhadi AM, Ramey W, et al. Revascularization and pediatric aneurysm surgery. *J Neurosurg Pediatr* 2014; 13(6): 641–646.
12. Buis DR, van Ouwerkerk WJR, Takahata H, et al. Intracranial aneurysms in children under 1 year of age: a systematic review of the literature. *Childs Nerv Syst* 2006; 22(11): 1395–1409.
13. Lasjaunias P, Wuppalapati S, Alvarez H, et al. Intracranial aneurysms in children aged under 15 years: review of 59 consecutive children with 75 aneurysms. *Childs Nerv Syst* 2005; 21(6): 437–450.
14. Levy ML, Levy DM and Manna B. Pediatric cerebral aneurysm. In: *StatPearls* [Internet]. Treasure Island, FL: StatPearls Publishing, 2023.
15. Sorteberg A and Dahlberg D. Intracranial non-traumatic aneurysms in children and adolescents. *Curr Pediatr Rev* 2013; 9(4): 343–352.
16. de Aguiar GB, Ozanne A, Elawady A, et al. Intracranial aneurysm in pediatric population: a single-center experience. *Pediatr Neurosurg* 2022; 57(4): 270–278.
17. Garrido E, Metayer T, Borha A, et al. Intracranial aneurysms in pediatric population: a two-center audit. *Childs Nerv Syst* 2021; 37(8): 2567–2575.
18. Chen R, Zhang S, Guo R, et al. Pediatric intracranial distal arterial aneurysms: report of 35 cases. *Acta Neurochir (Wien)* 2018; 160(8): 1633–1642.
19. Morel S, Diagbouga MR, Dupuy N, et al. Correlating clinical risk factors and histological features in ruptured and unruptured human intracranial aneurysms: The Swiss AneuX study. *J Neuropathol Exp Neurol* 2018; 77(7): 555–566.
20. Frösen J, Piippo A, Paetau A, et al. Remodeling of saccular cerebral artery aneurysm wall is associated with rupture. *Stroke* 2004; 35(10): 2287–2293.
21. Waters KM, Rooper LM, Guajardo A, et al. Histopathologic differences partially distinguish syndromic aortic diseases. *Cardiovasc Pathol* 2017; 30: 6–11.
22. Keedy A. An overview of intracranial aneurysms. *Mcgill J Med* 2006; 9(2): 141–146.
23. Mohotti JE, Carter NS, Zhang VJW, et al. Neonatal intracranial aneurysms: case report and review of the literature. *J Neurosurg Pediatr* 2018; 21(5): 471–477.
24. Kakarla UK, Beres EJ, Ponce FA, et al. Microsurgical treatment of pediatric intracranial aneurysms. *Neurosurgery* 2010; 67(2): 237–250.
25. Lauzier DC, Osbun JW, Chatterjee AR, et al. Safety of pediatric cerebral angiography. *J Neurosurg Pediatr* 2022; 29(2): 192–199.

26. Wilson JL, Eriksson CO and Williams CN. Endovascular therapy in pediatric stroke: utilization, patient characteristics, and outcomes. *Pediatr Neurol* 2017; 69: 87–92.e2.
27. Hoffman CE, Santillan A, Rotman L, et al. Complications of cerebral angiography in children younger than 3 years of age. *J Neurosurg Pediatr* 2014; 13(4): 414–419.
28. Sporns PB, Strater R, Minnerup J, et al. Feasibility, safety, and outcome of endovascular recanalization in childhood stroke: the Save ChildS study. *JAMA Neurol* 2020; 77(1): 25–34.
29. Riyaz M, Waqas M, Ujjan BU, et al. Decompressive craniectomy for infants: a case series of five patients. *Childs Nerv Syst* 2015; 31(11): 2117–2122.
30. Behbahani M, Rosenberg DM, Rosinski CL, et al. Cranioplasty in infants less than 24 months of age: a retrospective case review of pitfalls, outcomes, and complications. *World Neurosurg* 2019; 132: e479–e486.
31. Roque DA. Segmented versus nonsegmented deep-brain stimulation for essential tremor differ in ataxic side effects. *Tremor Other Hyperkinet Mov (N Y)* 2019; 9: 621.