A case of traumatic intracranial aneurysm in a 6-year-old child following a closed blunt head injury

Mariz Kaye Sales, MD[1]; Christopher Concepcion, MD, FAFN[1]; Asis Encarnacion, MD, FAFN[1]

Abstract

This is a case of a 6-year-old male who had a motor vehicular accident, who initially presented without neurologic deficit immediately post injury but followed up with seizures and motor weakness. The aim of the study is to review the pathomechanism of traumatic aneurysm in blunt head trauma in pediatric patients.

The initial imaging done revealed a closed, depressed comminuted fracture in the left frontal bone. Three weeks post-injury, he had recurrent generalized tonic clonic seizures and angiogram showed saccular aneurysm of the left A2 with left frontal intracerebral hemorrhage (ICH).

The patient underwent bifrontal craniotomy, clipping of aneurysm and evacuation of the ICH. Treatment options includes endovascular approach, wrapping and trapping the aneurysm. However, there is no single modality indicated for all lesions. Post-operatively, the patient was aphasic with right hemiparesis (2/5). Speech improved the second week after the surgery. Rehabilitation was initiated after admission and unassisted ambulation noted after 2 months. Work up done for other possible causes of aneurysm in the pediatric population revealed unremarkable findings. Mechanism associated with blunt traumatic head injury in the development of aneurysm could be secondary to a shear or rotational injury damaging vessels in close proximity to dura, for instance in this case, close to the falx cerebri.

Key words: Traumatic pediatric aneurysm, blunt head injury

The aim of the study was to review the pathomechanism of traumatic aneurysm in a blunt head trauma in a pediatric patient. As cited by Emir, et al., traumatic intracranial aneurysms (TICA) following closed head injury comprise less than 1% of all cerebral aneurysms and is associated with significant morbidity and mortality rate as high as 50%. They may present with headache acutely or delayed and may even be seen as an incidental finding in imaging studies.[1] A subarachnoid hemorrhage is the most common imaging finding of intracranial aneurysm in both pediatrics and adults. Similar to adult counterparts, pediatric aneurysms can be related to systemic diseases and/or other intracranial pathologies. Other systemic conditions may include Marfan Syndrome, polycystic kidney disease, coarctation of the aorta, and fibromuscular dysplasia. As for adults – acquired conditions such as diabetes mellitus, hypertension, cigarette smoking and hyperlipidemia, are noted risk factors for developing intracranial aneurysm. Pediatric intracranial aneurysm is considered rare and a threatening

Correspondence:
Mariz Kaye Sales, MD
Email: masales@uerm.edu.ph
[1]University of the East Ramon Magsaysay Memorial Medical Center

condition. Mortality is high if left untreated and sudden death may ensue with rebleeding. Management approach to these pediatric aneurysms largely differ from that of adults. The aim of the study was to review the pathomechanism of traumatic aneurysm in blunt head trauma in pediatric patients.

The Case

A consent was obtained from the father regarding presentation of the case.

This case involved a 6-year-old male who met developmental milestones prior to the injury and has no known familial comorbidities or previous hospitalizations. Before the injury, the patient excelled academically, shown proficiency in grammar, could read up to ten simple words with clarity, was capable of writing letters and numbers, and could ride a bicycle. He had a motor vehicular accident and following an emergency department consultation, exhibited a favorable condition immediately after the injury. The patient presented with a Glasgow Coma Scale score of 15, exhibiting alertness and no sensory or motor abnormalities; hence, he was monitored and discharged from the emergency room on the same day. At follow up after 4 days, he presented with recurrent seizures and motor weakness. Plain cranial computerized tomography (CT) scan was performed revealing a closed and depressed comminuted fracture in the left frontal bone. He was then admitted for observation and was discharged in apparent good health, with no recurrence of seizures. Eight days post injury, he complained of intermittent bifrontal to diffused headaches. A repeat plain cranial CT scan done revealed minimal interhemispheric subarachnoid hemorrhage, not previously seen on his prior CT scan. Patient was recommended for closed observation but was lost to follow up.

Three weeks post-injury, he had recurrent generalized tonic clonic seizures preceded by severe headache and decreased sensorium necessitating intubation during the emergency room consultation. Post ictus, the patient had eye opening to vigorous tapping, pupils were isocoric and briskly reactive to light, and with right hemiparesis. A cranial CT angiogram scan showed a saccular aneurysm of the left A2 branch aneurysm measuring 0.4cm × 0.4cm with its base measuring 0.27cm in its widest and left frontal intracerebral hemorrhage (ICH) measuring approximately 48 ml in volume (Figure 1 A and B)

The patient underwent bifrontal craniotomy, evacuation of the ICH and clipping of ruptured aneurysm. Post-operatively, the patient remained intubated, noted with right hemiparesis (2/5), was treated for pneumonia and weaned slowly. Post-extubation, the patient was aphasic until the second week after the surgery where the patient was able to speak a few words. Rehabilitation was initiated during admission and the patient was able to ambulate without support after 2 months and speech was comprehensible. Work-up for other possible causes of aneurysm in the pediatric population such as 2D echocardiography, whole abdominal ultrasound and hematologic work up all revealed unremarkable findings.

Discussion

Childhood aneurysms commonly become symptomatic at two periods - from birth to 6 years old and then from 8 years old to adolescence.[2] More commonly, cases seen are within the first 2 years of life although

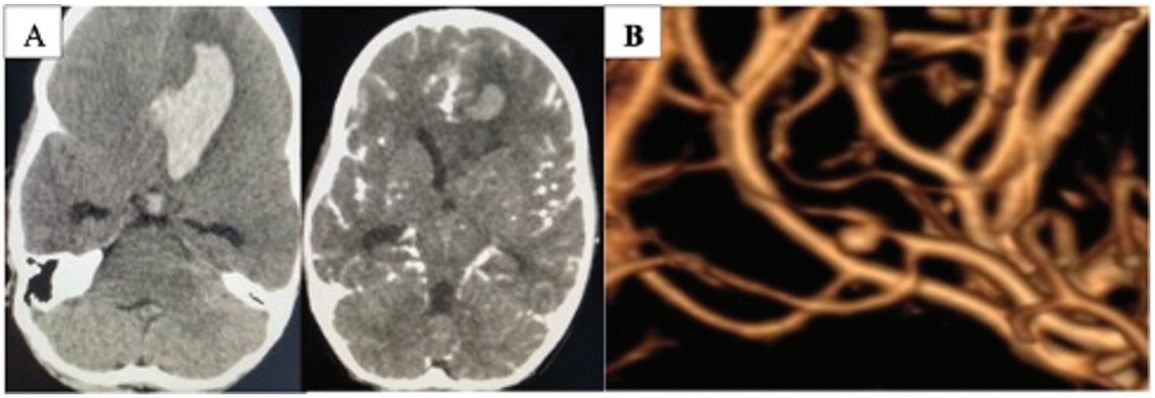

Figure 1. (A) Left frontal intracerebral hemorrhage (B) Saccular aneurysm at the left anterior cerebral artery, A2 segment

the early childhood age group includes newborns to the first 6 months. The incidence in children varies from 1.9% to 4.6%. Increasing trend of diagnosed cases is probably due to better diagnostic tools and heightened clinical suspicion.[1] Traumatic aneurysms can be classified as direct, if from a penetrating injury, or indirect in cases when it is caused by a blunt or closed head injury. The cortical vessels can be involved directly below or near the fractured segments.[2] Some cerebral aneurysms may be secondary to inherited risk factors such as genetic connective tissue disorders that weaken artery walls, polycystic kidney disease, arteriovenous malformations that disrupt blood flow and a history of aneurysm in a first degree family member. Traumatic intracerebral aneurysms are relatively common in childhood in the first 21 days post-trauma or even later.[3] Pediatric aneurysms can also be related to a systemic disease such as atherosclerosis, infection, connective tissue disease, familial syndromes or vascular anomalies due to unrecognized factors during development.[4,5,6] A higher incidence among patients with sickle cell disease was also noted.[5]

There are several theories postulating development of pediatric aneurysms. Traumatic aneurysms usually arise at the skull base or at distal anterior or middle cerebral arteries secondary to direct mural injury or to acceleration-induced shear.[6] They may also develop from the remnants of vascular trunks arising from arterial bifurcations.[2] These saccular aneurysms may arise from the vessel stumps that disappeared when fetal vascular network matured forming into the adult major arterial supply. These vestigial nubbins may form aneurysms over time. On the other hand, a degenerative process can also explain aneurysm development in the pediatric population. Intrinsic defects are considered as causative event predisposing vessel wall disruption hence the failure of repair on these subsequent insults. There are components of both intrinsic defects and insults in the development of aneurysmal disease amongst pediatric population.[2] A balance between these two factors and the compensation and repair for these changes can predict which aneurysm grows, ruptures, or regresses. Iatrogenic aneurysms may also occur with unexpected frequency in childhood and adolescence and they may also present early or late. Hence, any child with head injury who presents with delayed neurologic deterioration should undergo diagnostic imaging.[7] Pediatric intracranial aneurysms differ in comparison to adults in terms of characteristics, presentation and treatment outcomes.[8]

Endothelin receptor type A (EDNRA) gene encodes a receptor activated by endothelin which are responsible for the constriction and dilation of blood vessels. Endothelin-1, which is the primary variant present in vascular smooth muscle cells, is the one triggering EDNRA.[9] This signaling pathway is activated in vascular injuries causing increased in cell proliferation. There is a potential that a diminished activity, or downregulation, of EDNRA signaling might be a precursor to compromised vascular repair mechanisms. When this repair process is not optimized, it could cause vasculature susceptible to aneurysm formation following injuries or disturbances.[10]

In 15 to 25% of children with SAH, focal or generalized seizures can occur and are more common in infants. Seizure rate as the first presenting sign is about 7%. Seizures occurred most frequently in children with giant (25%) and posterior circulation (27%) aneurysms.[2] Once the presence of subarachnoid hemorrhage has been established, a standard, four-vessel cerebral angiogram is suggested for further investigation.

Traumatic aneurysms can be histologically categorized as true, false, or mixed. The most common type is false aneurysms. These aneurysms are typically associated with an acute episode of delayed intracranial hemorrhage with an average time from initial trauma to aneurysm hemorrhage of approximately 21 days.[3] Several mechanisms have been proposed in the formation of traumatic aneurysms, either direct injury to the vessel or via stretching of the vessel closely related to the anatomical location involved by adjacent forces. Infraclinoid carotid and basilar artery aneurysms are commonly associated with basilar skull fractures due to the proximity of these vessels with the skull base.[11,12,13]

The management of pediatric aneurysms is complex and difficult due to their distinct characteristics in contrast to adult cerebral aneurysms. A male predominance was seen, with the internal carotid artery bifurcation being the most prevalent site for the aneurysms.[8] In comparison to adults, pediatric patients exhibit a higher incidence of both posterior circulation and large aneurysms.[14]

Despite a higher incidence of complex aneurysm cases in pediatric patients, there is a good recovery at 6-months follow-up, which was seen in this case.

Post-operatively, the patient was aphasic with right hemiparesis (2/5). Speech improved the second week after the surgery and he was able to ambulate unassisted after 2 months. A favorable outcome of up to 95% is observed following the treatment of both ruptured and unruptured pediatric aneurysms.[15] The endovascular technique, wrapping, and trapping the aneurysm are further therapy possibilities. Nonetheless, no single approach is recommended for all lesions. The recurrence rate of treated aneurysms was 6.7% in the endovascular cohort and 0% in the microsurgical cohort. No de novo aneurysms developed in children, and the pediatric mortality rate was 10.6%.[16]

Conclusion

Traumatic Intracranial aneurysms (TICAs) may progressively grow, cause ICH and deterioration if not detected early. Hence high index of suspicion and aggressive treatment when diagnosed is generally needed. Post-operatively, the patient was aphasic with right hemiparesis (2/5). Speech improved the second week after the surgery. Rehabilitation was initiated after admission and unassisted ambulation noted after 2 months. Work up done for other possible causes of aneurysm in the pediatric population revealed unremarkable findings. Mechanism associated with blunt traumatic head injury in the development of aneurysm could be due to a shear or rotational injury damaging the vessels in close proximity to the dura, for instance in this case, close to falx cerebri.

References

1. Emir AF, Udin N, Othman AK, Salim NN, Saiful Arifin A, Rahman MM, Mohd R. Traumatic intracranial aneurysm (TICA) following closed head injury in children: a case report. JSM Clin Case Rep 2022 Nov;10(2):1204. Available from: https://doi.org/10.47739/2373-9819/1204.
2. Levy ML, Levy DM, Manna B. Pediatric cerebral aneurysm. Updated 2023 Aug 8. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024 Jan–. Available from: https://www.ncbi.nlm.nih.gov/books/NBK537085/.
3. Livshits IM, Berdinov BF, Musa G, Magomedov I, Askerkhanov A. Traumatic intracranial aneurysms (TICA) in children: a description of two clinical cases of successful treatment and review of literature. Childs Nerv Syst 2022 Nov;38(11):2063–70. Available from: https://doi.org/10.1007/s00381-022-05647-9.
4. Vaicys C, Hunt CD, Heary RF. Ruptured intracranial aneurysm in an adolescent with Alport's syndrome—a new expression of type IV collagenopathy: case report. Surg Neurol 2000 Jul;54(1):68–72. Available from: https://doi.org/10.1016/s0090-3019(00)00253-6.
5. Holmin S, Ozanne A, Zhao WY, Alvarez H, Krings T, Lasjaunias P. Association of cervical internal carotid artery aneurysm with ipsilateral vertebrobasilar aneurysm in two children: a segmental entity? Childs Nerv Syst 2007 Jul;23(7):791–8. Available from: https://doi.org/10.1007/s00381-006-0294-1.
6. Goia A, Garrido E, Lefebvre M, Agid R, Wang H, Wang F. Ruptured intracranial aneurysm in a neonate: case report and review of the literature. World Neurosurg 2020 Aug;140:219–23. Available from: https://doi.org/10.1016/j.wneu.2020.05.018.
7. Ventureyra EC, Higgins MJ. Traumatic intracranial aneurysms in childhood and adolescence: case reports and review of the literature. Childs Nerv Syst 1994 Aug;10(6):361–79. Available from: https://doi.org/10.1007/BF00335125.
8. Kim M, Lee HS, Lee S, Park JC, Ahn JS, Kwon DH, Kwun BD, Park W. Pediatric intracranial aneurysms: favorable outcomes despite rareness and complexity. World Neurosurg 2019 May;125:e1203–16. Available from: https://doi.org/10.1016/j.wneu.2019.01.280.
9. Wang X, Douglas SA, Louden C, Vickery-Clark LM, Feuerstein GZ, Ohlstein EH. Expression of endothelin-1, endothelin-3, endothelin-converting enzyme-1, and endothelin-A and endothelin-B receptor mRNA after angioplasty-induced neointimal formation in the rat. Circ Res 1996 Feb;78(2):322–8. Available from: https://doi.org/10.1161/01.res.78.2.322.
10. Khatri D, Zampolin R, Behbahani M, Kobets A, Lax D, Manwani D, Benitez S, Toma A, Holland R, Brook A, Lee SK. Pediatric brain aneurysms: a review of 1458 brain MR angiograms. Childs Nerv Syst 2023 Nov;39(11):3249–54. Available from: https://doi.org/10.1007/s00381-023-05967-4.
11. Bank WO, Nelson PB, Drayer BP, Wilkins RH, Rosenbaum AE. Traumatic aneurysm of the basilar artery. AJR Am J Roentgenol 1978 May;130(5):975–7. Available from: https://doi.org/10.2214/ajr.130.5.975.
12. Loop JW, White LE Jr, Shaw CM. Traumatic occlusion of the basilar artery within a clivus fracture. Radiology 1964 Jul;83:36–40. Available from: https://doi.org/10.1148/83.1.36.
13. Maurer JJ, Mills M, German WJ. Triad of unilateral blindness, orbital fractures, and massive epistaxis after head injury. J Neurosurg 1961 Nov;18:837–40. Available from: https://doi.org/10.3171/jns.1961.18.6.0837.
14. Mehrotra A, Nair AP, Das KK, Srivastava A, Sahu RN, Kumar R. Clinical and radiological profiles and outcomes in pediatric patients with intracranial aneurysms. J Neurosurg Pediatr 2012 Oct;10(4):340–6. Available from: https://doi.org/10.3171/2012.7.PEDS11455.
15. Huang J, McGirt MJ, Gailloud P, Tamargo RJ. Intracranial aneurysms in the pediatric population: case series and literature review. Surg Neurol 2005 May;63(5):424–32; discussion 432–3. Available from: https://doi.org/10.1016/j.surneu.2004.11.023.
16. Xu R, Xie ME, Yang W, Gailloud P, Caplan JM, Jackson CM, Jackson EM, Groves ML, Robinson S, Cohen AR, Huang J, Tamargo RJ. Epidemiology and outcomes of pediatric intracranial aneurysms: comparison with an adult population in a 30-year, prospective database. J Neurosurg Pediatr 2021 Sep 10;28(6):685–94. Available from: https://doi.org/10.3171/2021.6.PEDS21268.