SUCCESSFUL ENDOVASCULAR MANAGEMENT OF A RUPTURED CEREBRAL ANEURYSM IN AN INFANT PATIENT

Manejo endovascular exitoso de un aneurisma cerebral roto en un paciente lactante

JOHN VARGAS U.[1a], JESÚS FLORES Q.[1b], RODOLFO RODRÍGUEZ V.[1b], WALTER DURAND C.[1b], DANTE VALER G.[1b]

[1]Service of Neuroradiology, Department of Neurosurgery of the Guillermo Almenara National Hospital, Lima, Peru.
[a]Resident of Neurosurgery, [b]Endovascular Neurosurgeon

ABSTRACT

Introduction: Cerebral aneurysms in pediatric age are rare. In early childhood they appear before the age of 2 years and are related to a high incidence of injuries along the middle cerebral artery, in its distal part and in the vertebrobasilar system. The etiology can be idiopathic, traumatic, and fungal. Aneurysm obliteration should be as early as possible in patients with low surgical risk.
Clinical Case: The case of a 5-month-old patient with no significant medical history is presented, with a 2-day illness time, signs of irritability, vomiting and tension in the fontanelle. A cerebral tomography was performed showing a predominantly right subcortical frontal fine subarachnoid hemorrhage and an angio-tomography (Angio-TEM) that showed an aneurysm of the anterior cerebral artery. The cerebral angiography study revealed a dissecting aneurysm of the left A2-A3 segment that involved the division of the left anterior cerebral artery into a pericallosal and marginal callus artery. An embolization was performed using 4 coils and Histoacryl® to close the parental artery. He had a seizure crisis from a left marginal callus infarction that was medically controlled. The clinical evolution was good, being discharged on the 7th day of hospitalization.
Conclusion: Pediatric cerebral aneurysms are a rare pathology and in patients with low surgical risk, such as our patient, they should be treated as soon as possible to decrease morbidity and mortality.

Keywords: *Intracranial Aneurysm, Cerebral Angiography, Infant, Embolization Therapeutic. (Source: MeSH NLM)*

RESUMEN

Introducción: Los aneurismas cerebrales en edad pediátrica son muy raros. En la infancia temprana se presentan antes de los 2 años y están relacionados con alta incidencia de lesiones a lo largo de la arteria cerebral media, en su parte distal y también en el sistema vertebro-basilar. La etiología puede ser idiopática, traumática y micótica. La obliteración del aneurisma debe ser lo más temprano posible en los pacientes con bajo riesgo quirúrgico.
Caso Clínico: Se presenta el caso de una paciente de 5 meses de edad, sin antecedentes médicos de importancia, con tiempo de enfermedad de 2 días, signos de irritabilidad, vómitos y tensión en la fontanela. Se le realizó una tomografía cerebral donde se evidenció una hemorragia subaracnoidea fina frontal subcortical a predominio derecho y una Angiotomografía (AngioTEM) que mostró un aneurisma de la arteria cerebral anterior. El estudio de angiografía cerebral evidenció un aneurisma disecante del segmento A2-A3 izquierdo que involucraba la división de la arteria cerebral anterior izquierda en arteria pericallosa y callosa marginal. Se realizó una embolización utilizando 4 coils e Histoacryl® para cerrar la arteria parental. Presentó una crisis convulsiva a causa de un infarto calloso marginal izquierdo que se controló médicamente. La evolución clínica fue buena siendo dado de alta al 7mo día de hospitalización.
Conclusión: Los aneurismas cerebrales pediátricos son una patología rara y en pacientes con bajo riesgo quirúrgico como nuestra paciente deben ser tratados a brevedad posible para disminuir la morbimortalidad.

Palabras Clave*: Aneurisma Intracraneal, Angiografía cerebral, Lactante, Embolización Terapéutica. (Fuente: DeCS Bireme)*

Peru J Neurosurg 2020, 2 (2): 55-60

Cerebral aneurysms in pediatric age are very rare, being the first case described in 1871 by Eppinger in a necropsy of a 15-year-old patient. It is known that, in adult patients, the formation of cerebral aneurysms is associated with diabetes, alcohol, smoking, obesity, use of oral contraceptives and high blood pressure, but the same does not occur in children, which is why many authors give more importance to genetic factors in the development of aneurysms in the pediatric age. [1, 2]

Submitted *: March 21, 2020*
Accepted *: April 20, 2020*
HOW TO CITE THIS ARTICLE: Vargas J, Flores-Q J, Rodríguez R, Durand W, Valer D, Saal-Zapata G. Successful endovascular management of an unruptured cerebral aneurysm in an infant patient. *Peru J Neurosurg 2020; 2(2): 55-60*

The incidence of pediatric aneurysms is estimated at 1 to 3 cases per million people. In childhood, they appear more before the age of 2 years and are related to a high incidence of injuries along the middle cerebral artery in its distal part and in the vertebrobasilar system. Furthermore, pediatric aneurysms have been found to be more frequent in males, with a ratio of 1.3 to 2.8: [1], and it has also been noted that up to 62% of giant aneurysms occur in males.[1, 3]

The treatment of aneurysms at this age can be performed both surgically and endovascularly, however surgical treatment may carry a higher risk because blood loss is an important limitation at this age, hence the clipping of the aneurysm only occurs in 29.5% of cases. For this reason, a high percentage of pediatric patients require innovative procedures to close these lesions.[1, 4]

We present the case of a 5-month-old female infant with a dissecting aneurysm of the territory of the anterior brain (segment A2-A3) who was successfully treated endovascularly at the Neuroradiology Service of the Guillermo Almenara Hospital.

CLINICAL CASE

History and examination: 5-month-old female lactating patient from Lima, with no significant medical history. The illness time was 2 days characterized by irritability and vomiting of milk content on 10 occasions, so she was brought to the hospital emergency. On examination, the patient presented a greater tendency to sleep, Glasgow Coma Scale (GCS): 14, without motor deficit, isochoric and reactive pupils, slightly tense anterior fontanel. Encephalitis is suspected, for which they perform a non-contrast brain tomography (CT) showing subcortical cortical subarachnoid hemorrhage at the bilateral frontal level, predominantly on the right. A cerebral angiotomography (angioCT) is performed, showing a dysplastic aneurysm that depends on segment A2 of the right anterior cerebral artery (*Fig 1*).

Endovascular treatment: A cerebral panangiography was performed on the third day of admission, a 4Fr introducer was placed in the right femoral artery, under the Seldinger technique, then with a Fargo Mini 4.2Fr ® guide catheter assisted with the Poseidon 150cm ® hydrophilic guide. An angiography of bilateral internal carotid artery (ICA) and left vertebral artery in anteroposterior, lateral, and oblique incidences, as well as 3D study of left ICA, where a dysplastic, dissecting aneurysm of left A2-A3, involving division of the left anterior cerebral artery (ACA) in the pericallosal and marginal callus artery (*Fig 2*), 13.67x4.57 mm.

The aneurysm was then embolized, for which the Headway Duo ® microcatheter assisted with the Traxcess 14 ® microguide was navigated until the aneurysm dome itself was reached through the left ACA, where it was embolized with coils, first with an Axium Prime ® 6mmx10cm, then a Microplex 10 ® 4mmx12cm, followed by a Microplex 10® 4mmx12cm and a fourth fourth Microplex 10 ® coil 3mmx8cm (*Fig 3*).

Due to the pathophysiology of the origin of this aneurysm, it was decided to close the artery from which the aneurysm originated to avoid recanalization, so it was embolized with 0.2mL of a dilution of Histoacryl ® with Lipiodol ® of 1: 2, managing to close the left marginal callus artery. Angiographic controls were performed showing the Raymond Roy I embolized aneurysm (*Fig 4*).

Clinical evolution: The patient was transferred to the pediatric intensive care unit (ICU) where she remained with GCS: 15 points, without motor or sensory deficit, isochoric and photoreactive pupils, unchanged cranial nerves, and slightly tense fontanel. A CT scan of the brain without contrast was performed, showing embolization material in the territory of the left ACA, without abnormal findings.

On the fourth postoperative day, the patient presented a seizure crisis, and in the CT scan of the brain without contrast, an infarct of the territory of the left marginal callus

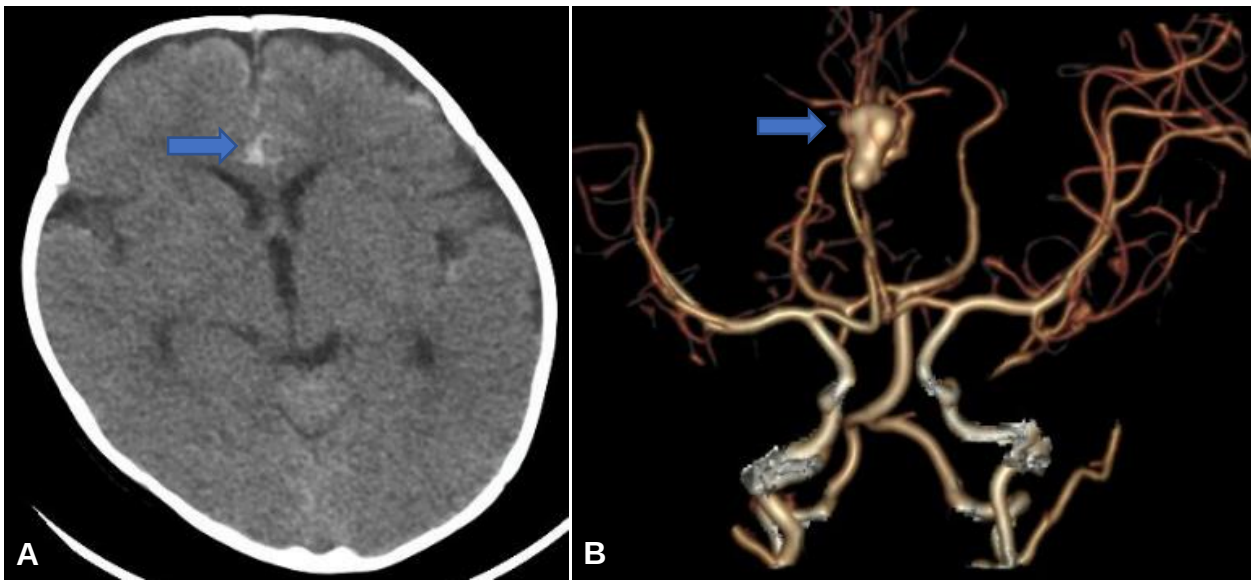

Fig 1. (A) Brain CT without contrast in axial section showing subarachnoid hemorrhage predominantly subcortical at the interhemispheric bifrontal level, predominantly on the right side (*blue arrow*). **(B)** 3D reconstruction of cerebral Angiotomography, showing dysplastic aneurysm, at the level of the marginal callus arteries (*blue arrow*)

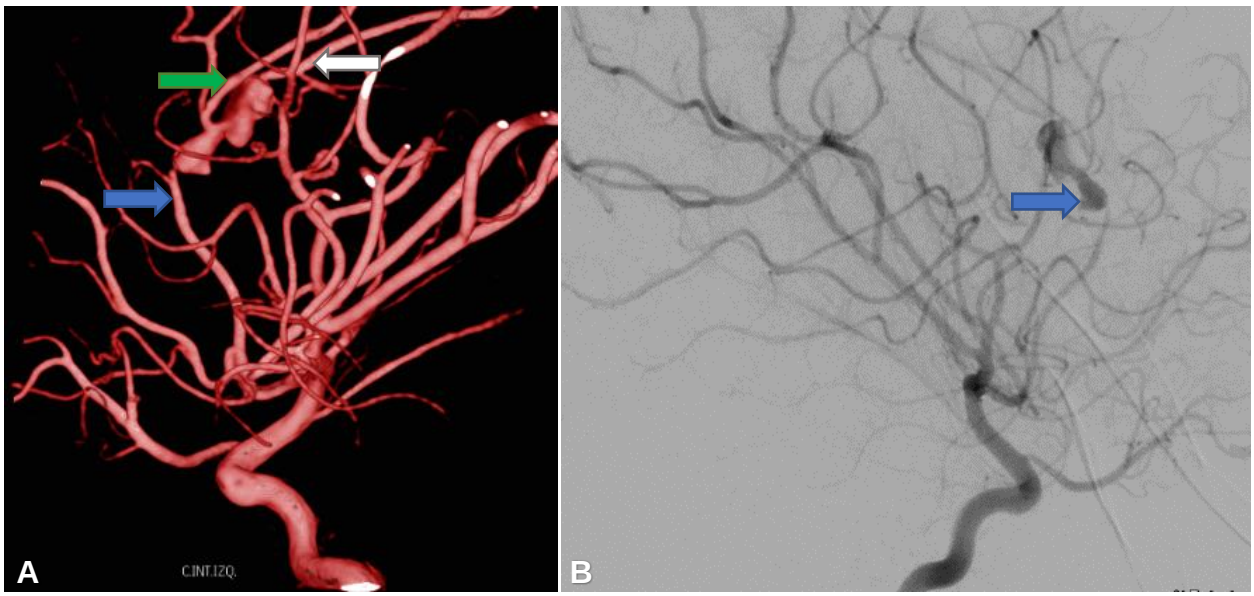

Fig 2. (A) 3D angiography of the left ICA showing a dissecting aneurysm at the bifurcation of the anterior cerebral artery (blue arrow) in the pericallosal artery (white arrow) and the marginal callus artery (green arrow). **(B)** Digital subtraction angiography in lateral view showing dissecting aneurysm of the left anterior cerebral artery (blue arrow)

artery was observed, but without clinical evidence of motor deficit (*Fig 5*). The patient continued with good clinical evolution, without seizures, so she was discharged on day 7 postoperatively.

DISCUSSION

Brain aneurysms in children are a rare pathology, being more frequent below 2 years.[1] The most common location is the carotid bifurcation, which can be found in 29 to 54% of cases, compared to adults who are from 4.4 to 4.5%. Likewise, the presence of multiple aneurysms in children is rarer (4-5%) than in adults (15%), and if they were present they would be related to cranial irradiation, Moya-Moya disease, sickle cell anemia, myxoma cardiac, AVM, or fibromuscular dysplasia. [1, 4]

Regarding the etiology, pediatric aneurysms can be of idiopathic, traumatic or infectious etiology (fungal aneurysms). There is much controversy about the idiopathic nature of aneurysms, but cadaveric studies show fragmentation of the internal elastic membrane and the medial muscle layer. In the aneurysm dome, the muscle layer is absent, but the internal elastic membrane continues along the neck of the lesion. Furthermore, the basement membrane under the endothelium is often thin and reticulated. The site of rupture of the aneurysm is at its apex and mimics a small diverticulum, very similar to adult saccular aneurysms. [2,4]

Aneurysms in childhood have been associated with various vascular diseases of collagen and connective tissue, where type I and type III collagen are known to predominate in the intracranial circulation and provide greater support to blood vessels. Ostergaard postulates that deficiency of type III collagen causes increased compliance of the blood vessel wall, hence a greater tendency to larger size in childhood aneurysms.[1] Atheromatous degeneration and hemodynamic factors are factors associated with aneurysm formation, but these factors are rare in children. Arterial hypertension in children can be seen in association with aortic coarctation and polycystic kidney disease, so if they coexist, the possibility of this entity as an etiological agent should not be ruled out. [3, 5]

In patients with a history of closed head trauma, traumatic damage to the blood vessel wall can lead to the formation of an aneurysm. These types of aneurysms have specific locations, such as the anterior cerebral artery and its distal branches due to damage along the sickle, the posterior cerebral artery near the free edge of the tentorium, the middle cerebral artery along its course through the sphenoid wing. . True traumatic aneurysms have a single intact layer of adventitia without following the classic layers of disruption. False traumatic aneurysms result from a fibrous organization around a primary hematoma that surrounds a parental vessel, which can develop in cases of depressed skull fractures or penetrating injuries. [2, 4]

The term fungal was originally given by Osler in 1885, but most infectious aneurysms in childhood are given by bacteria rather than fungi. The fungi most involved in the development of aneurysms were Aspergillus, Candida and Phycomycetes, in descending order of frequency. These patients are generally immunocompromised and had a mortality of close to 100%. Most commonly, it is by hematogenous spread from endocarditis, but may also be by direct extension from an adjacent nest of infection. The bacteria involved were alpha streptococcus, staphylococcus, pseudomonas, and Hemophilus. The time from the embolic event to the formation of the aneurysm is very short, generally 2 days to hemorrhage according to the Molinari model. [1]

In our patient, the location of the ACA and distal aneurysm indicated that it was a traumatic aneurysm, although there was no history of closed head trauma.

In patients who have been diagnosed with subarachnoid hemorrhage (SAH), initial medical management is

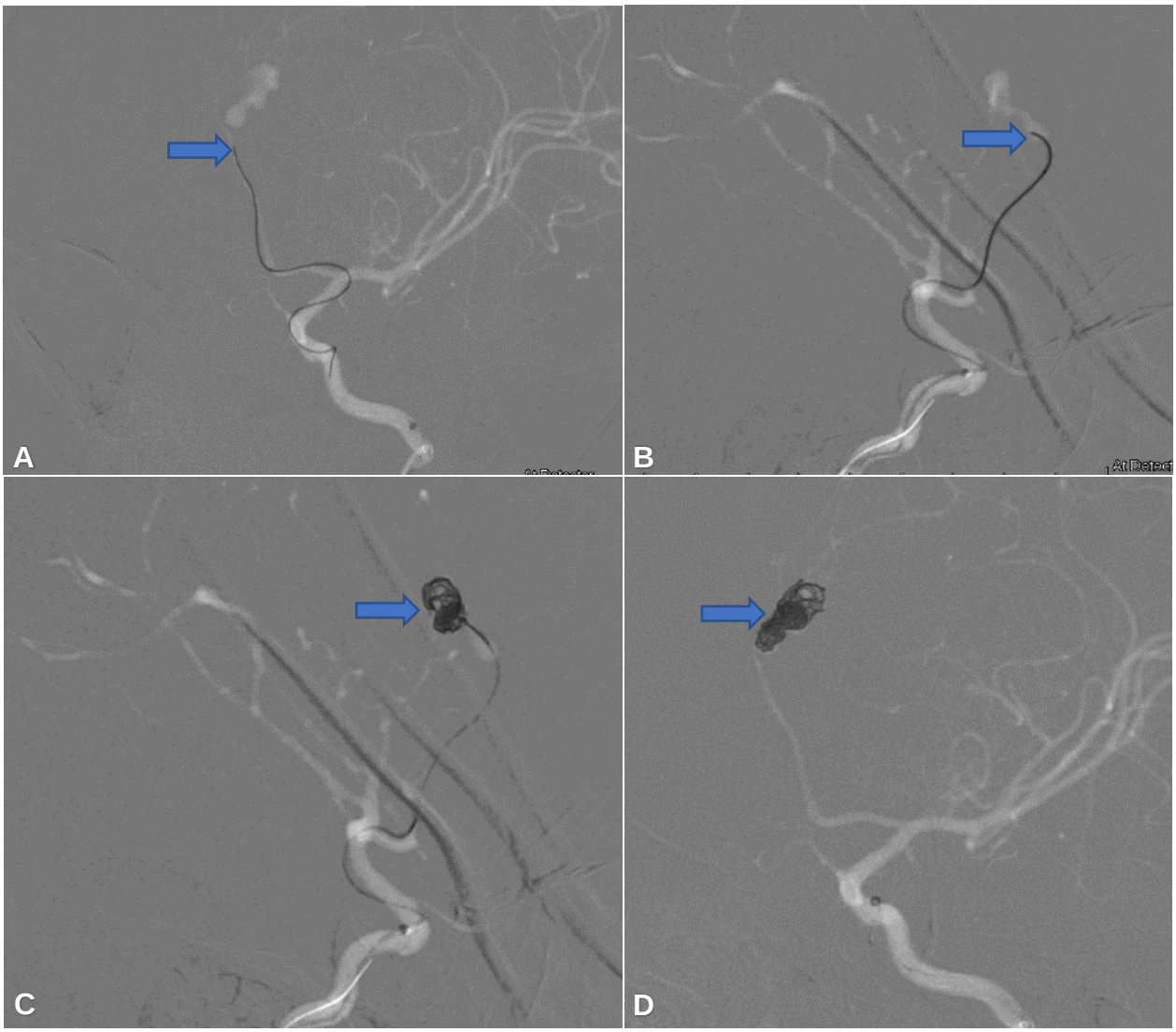

Fig 3. (A) Road mapping in anteroposterior incidence of the left ICA where evidence of advancement of the microcatheter and the microguide (arrow) through the left anterior cerebral artery is evident. **(B)** Road mapping in lateral incidence of the left ICA, where the advancement of the microguide to the aneurysm dome itself is evident (arrow). **(C)** Road mapping in lateral incidence of the left ICA in which the aneurysm is being embolized with a coil (arrow). **(D)** Road mapping in anteroposterior incidence of the left ICA after having embolized with all coils the aneurysm (arrow) and removal of the microguide.

important to stabilize the patient, since within the first 48 hours the most serious complication is rebleeding.

Placement of the peritoneal ventricle bypass (VPS) in hydrocephalus is indicated, but there is controversy whether to place it before treating the aneurysm, since this generates changes in transluminal pressure with possible rebleeding of the aneurysm due to it. [8, 9]

The use of nimodipine in children is limited, but in adults it has been shown to decrease the incidence of severe neurological deficit due to vasospasm. If clinical vasospasm is evident in children, it is preferable to use volume expansion, hyperdynamic therapy with vasopressors and inotropics. Other medical complications such as hyponatremia must also be prevented because it can exacerbate the deterioration of consciousness and increase epileptogenic activity. [11, 12]

It has been seen that spontaneous thrombosis of the aneurysm may exist in children, therefore conservative management is within the possibilities, but multiple studies have shown that these patients have a worse prognosis. This prognosis improves if a microsurgical or endovascular management is chosen as soon as possible. In cases of fungal aneurysms, conservative treatment with antibiotics may be reasonable, but surgical management may also be chosen. If the patient has multiple aneurysms with pre-existing heart disease and endocarditis, conservative management is best due to the high surgical risk. Aneurysms usually disappear after 2 to 3 weeks of medical treatment in these cases. [1]

Obliteration of the aneurysm should be as early as possible in patients with low surgical risk, but in those with high surgical risk, early treatment is controversial and it is preferred to wait 7 to 14 days after surgery of SAH, which allows greater stabilization of the sick patient, resolution of cerebral edema and vasospasm, thus reducing morbidity and mortality. [1, 3]

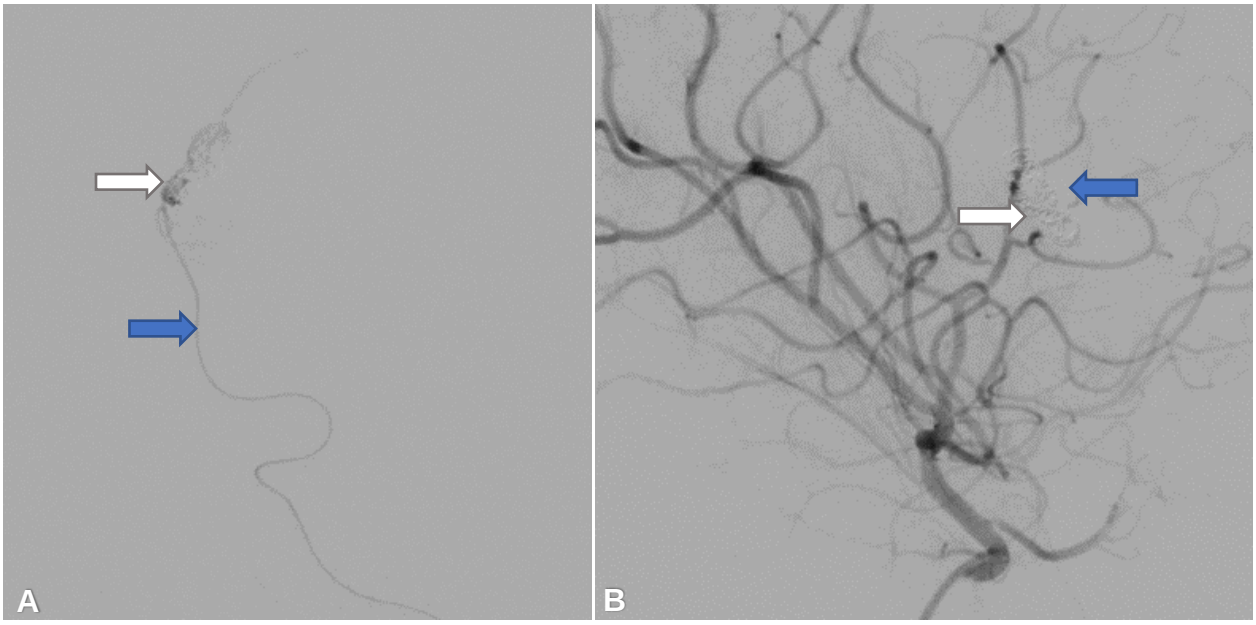

Fig 4. (A). Angiography in anteroposterior incidence of the left ICA, showing the path of the microcatheter (blue arrow) and Histoacryl embolization of the branch from which the dissecting aneurysm was born (white arrow). **(B)** Angiography in lateral incidence of the left ICA showing absence of the aneurysm (white arrow) and absence of the marginal callus artery (blue arrow)

In our patient, it was necessary to better recognize the characteristics of the aneurysm to proceed to treat it as soon as possible, and due to the small anatomical structures, the treatment was more complex. Microsurgical treatment was a possibility, but it had to be taken into account that blood loss in infants is crucial and can generate decompensation with small volumes of loss. Hence, the clipping of the aneurysm is only performed in 29.5% of cases. [1, 4]

Due to the aforementioned, endovascular management was chosen in our patient, and due to the pathophysiology, being a dissecting aneurysm, an artery had to be sacrificed. Fortunately, this did not produce major clinical repercussions since, as is known, children have neuronal neuroplasticity, which helps to improve the prognosis of these patients.[12, 13] The small infarction that occurred in the postoperative period was an epileptogenic focus, so the anticonvulsants were continued to avoid subsequent seizures in the girl, but despite this inconvenience, the patient had a good clinical course and was discharged within the of the first week of the event.

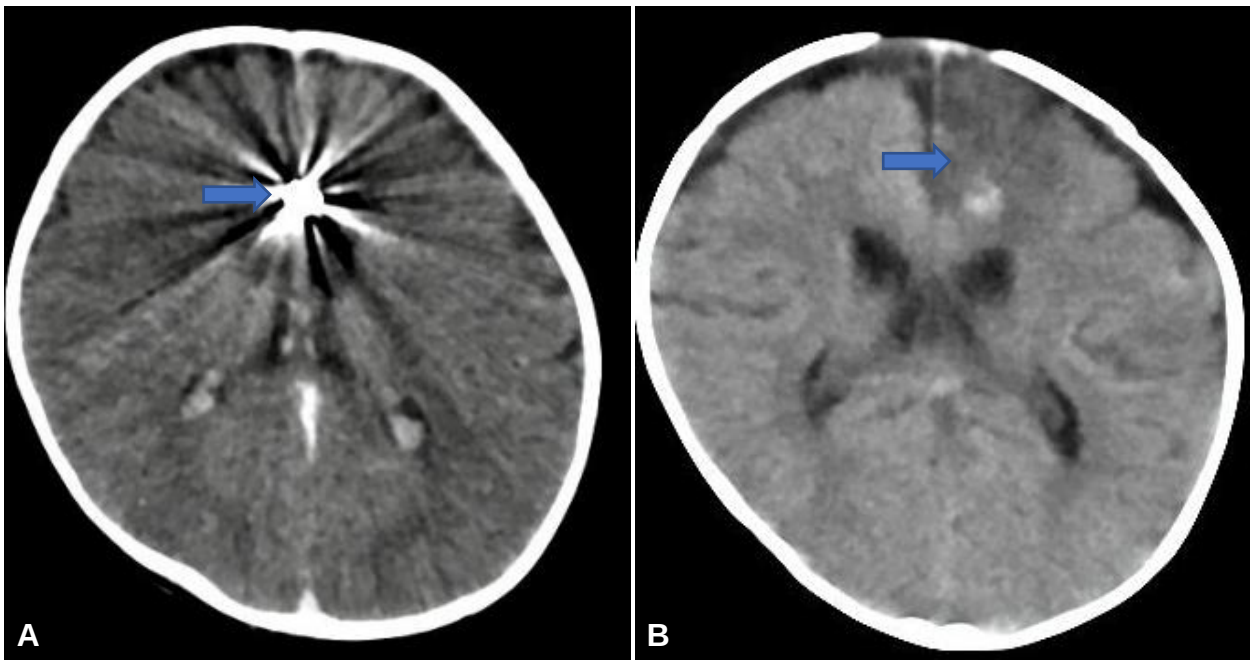

Fig 5. (A) Brain CT without contrast in axial section showing the hyperrefringence of the embolization material in the territory of the left A2-A3 segment (*blue arrow*). **(B)** Brain CT without contrast in axial section showing infarction in the territory of the left pericallosal artery (*blue arrow*) that did not cause major clinical repercussions.

CONCLUSION

Pediatric cerebral aneurysms are a rare pathology and it is necessary to determine their etiology to achieve adequate treatment. In patients with low surgical risk, the aneurysm should be treated as soon as possible, thus reducing the patient's morbidity and mortality, and this treatment can be better carried out endovascularly as this type of approach reduces hospital stay and the complications associated with it.

REFERENCES

1. Levy M, Levy D, Manna B. Pediatric cerebral aneurysm. StatPearls Publishing LLC. 2014; 12(1).
2. Aeron G, Abruzzo TA, Jones BV. Clinical and imaging features of intracranial arterial aneurysms in the pediatric population. **Radiographics. 2012** May-Jun;32(3):667-81.
3. Burke MJ. Occult aneurysmal hemorrhage in a child. Case report and literature review. **Pediatr Neurosurg. 2000** Nov;33(5):274-277.
4. Manz HJ, Vester J, Lavenstein B. Dissecting aneurysm of cerebral arteries in childhood and adolescence. Case report and literature review of 20 cases. **Virchows Arch a Pathol Anat Histol. 1979** Oct;384(3):325-35.
5. 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.
6. Beyens A, Albuisson J, Boel A, Al-Essa M, Al-Manea W, Bonnet D, Bostan O et Al. Arterial tortuosity syndrome: 40 new families and literature review. **Genet. Med. 2018 Oct;20**(10):1236-1245.
7. Ostergaard JR. Aetiology of intracranial saccular aneurysms in childhood. **Br J Neurosurg. 1991;5**(6): 575-80.
8. Kanaan I, Lasjaunias P, Coates R. The spectrum of intracranial aneurysms in pediatrics. **Minim Invasive Neurosurg. 1995** Mar;38(1):1-9.
9. Koroknay-Pál P, Niemelä M, Lehto H, Kivisaari R, Numminen J, Laakso A, Hernesniemi J. De novo, and recurrent aneurysms in pediatric patients with cerebral aneurysms. **Stroke. 2013 May;44**(5):1436-9.
10. Chen R, Zhang S, Guo R, Ma L, You C. Pediatric intracranial distal arterial aneurysms: report of 35 cases. **Acta Neurochir (Wien). 2018** Aug;160(8): 1633-1642.
11. Chen R, Zhang S, Guo R, You C, Ma L. Pediatric Intracranial Pseudoaneurysms: A Report of 15 Cases and Review of the Literature. **World Neurosurg. 2018** Aug;116: e951-e959.
12. Sujijantarat N, Pierson MJ, Kemp J, Coppens JR. Staged Trapping of Traumatic Basilar Trunk Pseudoaneurysm: Case Report and Review of Literature. **World Neurosurg. 2017 Dec;108**: 991.e7-991.e12.
13. Munakomi S, Tamrakar K, Chaudhary P, Bhattarai B, Cherian I. Case Report: Traumatic anterior cerebral artery aneurysm in a 4-year old child. **F1000Res. 2015**; 4:804.
14. Flores BC, Patel AR, Braga BP, Weprin BE, Batjer HH. Management of infectious intracranial aneurysms in the pediatric population. **Childs Nerv Syst. 2016 Jul;32**(7):1205-17.
15. Vávrova M, Jonszta T, Czerný D, Hrbac T, Lipina R, Klement P, Procházka V. Endovascular treatment of mycotic pseudoaneurysms. **VASA. 2010 Aug;39**(3): 256-61.

Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Authors Contributions
Conception and design: All authors. *Drafting the article:* Vargas. Critically revising the article: Rodriguez, Flores. *Reviewed submitted version of manuscript:* Vargas. *Approved the final version of the manuscript on behalf of all authors:* Vargas.

Correspondence
John Vargas Urbina. Department of Neurosurgery. Guillermo Almenara National Hospital. 800 Grau Avenue. La Victoria. Lima 13, Perú. *E-mail:* johnkilin27@hotmail.com