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SNI: Unique Case Observations

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Case Report

Long-term surveillance in an infant with spontaneous obliteration of pial arteriovenous malformation and large intranidal aneurysm: A unique case observation

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Received: 17 January 2024
Accepted: 23 May 2024
Published: 21 June 2024

DOI
10.25259/SNI_45_2024

Quick Response Code:

ABSTRACT

Background: Spontaneous obliteration of untreated cerebral arteriovenous malformations (AVMs) is rare, occurring in <1% of cases, and is even less common in pediatric populations. The mechanisms driving spontaneous regression of brain AVMs remain poorly understood, and long-term surveillance in pediatric patients is infrequently documented.

Case Description: The authors reported a remarkably rare instance of spontaneous thrombosis in a pial AVM accompanied by a large intranidal aneurysm in a 10-month-old infant, initially presenting with a nocturnal seizure. Diagnostic imaging revealed a ruptured intranidal aneurysm causing acute hemorrhage in the left anterior interhemispheric subdural space, extending into adjacent areas. Further, magnetic resonance imaging (MRI) and magnetic resonance angiography delineated the AVM in the left superior frontal gyrus, associated with a thrombosed aneurysm and surrounding edema. Cerebral angiography confirmed the AVM's origin from the left anterior cerebral artery, displaying early venous drainage and small, indirect feeders not amenable to endovascular treatment. Over time, serial imaging showed the aneurysm's transition from partial to complete thrombosis. Subsequent MRIs and angiographic assessments up to age 10 confirmed complete resolution of the AVM and aneurysm, with focal hyperemia persisted until age 16, when recurrent AVM was identified.

Conclusion: We document a rare spontaneous regression of a pial AVM with an intranidal aneurysm influenced by specific vascular factors. Despite this, spontaneous thrombosis should not replace vigilant long-term monitoring in pediatric neurovascular care.

Keywords: Pial Arteriovenous Malformation, Pediatric Neurovascular, Spontaneous Thrombosis, Focal hyperemia, Recurrent brain arteriovenous malformations, Intranidal aneurysm

INTRODUCTION

Brain arteriovenous malformations (AVMs) represent a complex and challenging neurovascular disorder, particularly in pediatric populations, and create a unique set of clinical challenges. These malformations are abnormal tangles of blood vessels and are characterized by direct connections between arteries and veins in the brain without an intervening capillary bed.[13,17] The

pathophysiology of brain AVMs in children is distinguished by its dynamic nature, influenced by the ongoing cerebral development and vascular remodeling inherent to this age group.[22]

The pial AVMs are a rare neurovascular anomaly in children under 2 years of age. These AVMs begin to resemble adult counterparts in their physiology, type, and architecture when children reach the age of 5–7 years.[19] In the broader demographic, including adults and a smaller proportion of children, the occurrence of spontaneous regression in brain AVMs is found to be remarkably low <1%.[1,5] Spontaneous thrombosis of pediatric AVMs, particularly in infants, represents an even rarer phenomenon.[11,20] Before our report, the literature has documented just one such instance in an infant.[23]

In 2019, we reported on an extremely rare case of a 10-month-old infant who experienced complete spontaneous thrombosis of a pial AVM associated with a large intranidal aneurysm.[14] This initial report detailed the acute phase and immediate management, highlighting the early spontaneous thrombosis and subsequent imaging findings up to the age of 11 years, which documented the complete obliteration of both the AVM and the aneurysm.

The present study extends the observations of this patient from now to 16 years of age, providing a unique long-term perspective on the natural history and neurodevelopmental outcomes of post spontaneous thrombosis of an AVM in a pediatric patient. This extended follow-up has allowed us to observe not only the stability of the thrombosis over a prolonged period but also to evaluate the long-term neurological and developmental impact of such rare spontaneous events in pediatric neurovascular disorders. Our findings contribute to a deeper understanding of the long-term clinical course and radiological evolution of pediatric AVMs, particularly those that undergo spontaneous resolution, and highlight the necessity for ongoing surveillance in such cases.

CASE DESCRIPTION

A 10-month-old female infant was admitted to a community hospital after experiencing a nocturnal seizure observed by her mother. The seizure involved screaming, eyes rolling up, and generalized convulsions lasting for 2 min, with no preceding fever or trauma. Born at term with a birth weight of 3080 g, her developmental history had been unremarkable. Initial diagnostic imaging through a computed tomography scan revealed a hyperdense, approximately 15 mm mass with surrounding edema in the left superior frontal gyrus and an adjacent acute thin subdural hematoma (SDH) [Figure 1]. Despite antiepileptic medication, the infant continued to experience seizures and required intubation and transfer to the intensive care unit. The provisional diagnosis was a ruptured aneurysm causing acute SDH, with a differential diagnosis of a hemorrhagic tumor.

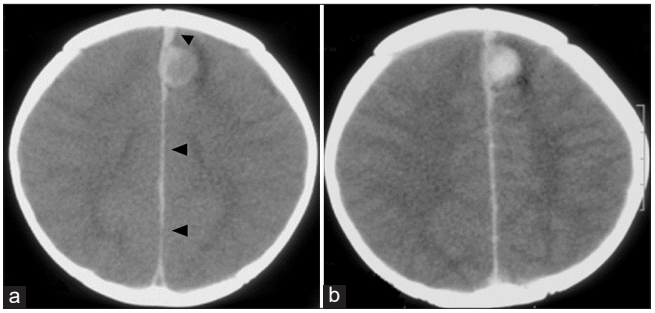

Figure 1: At the presentation. Axial views of cranial computed tomography scan (a) without and (b) with contrast show an enhancing hyperdense round mass, approximately 15 mm in size, with surrounding edema at the medial aspect of the left superior frontal gyrus. There was a small hyperdense lesion (black arrowheads) located at the left side of the anterior interhemispheric fissure adjacent to the medial part of the mass, extending posteriorly along the interhemispheric fissure, representing acute interhemispheric subdural hemorrhage.

A follow-up magnetic resonance imaging (MRI) obtained a week post symptom onset showed the SDH extending along the posterior interhemispheric fissure, posterior falx, bilateral tentorial cerebelli, and over both hemispheric convexities, more pronounced on the left side. Abnormal flow voids suggestive of a pial AVM were noted in the left superior frontal region. The left anterior interhemispheric SDH adjoined the medial side of the large round mass, indicating a probable source of hemorrhage from a ruptured aneurysm. T1-weighted gadolinium-enhanced MRI delineated the mass as a large, partially thrombosed aneurysm with eccentric signal voids and peripheral enhancement. T2-weighted MRI highlighted the progression of perianeurysmal edema in the same region. Magnetic resonance angiography (MRA) confirmed the pial AVM with a large intranidal aneurysm [Figure 2].

After a 3-week hospitalization, the infant was clinically stable and referred to our institute for potential endovascular treatment. Physical examination revealed mild developmental delay, closed anterior fontanel, and normal vital signs, with measurements of 44 cm head circumference, 7.5 kg weight, and 70 cm length. Neurological examination showed no signs of meningeal irritation, papilledema, or other neurological deficits.

One month after symptom onset, a follow-up MRI indicated complete resolution of the SDH and a laminated appearance in the partially thrombosed aneurysm, with persistent perianeurysmal edema [Figure 3]. Cerebral angiography confirmed a small pial AVM with a large intranidal

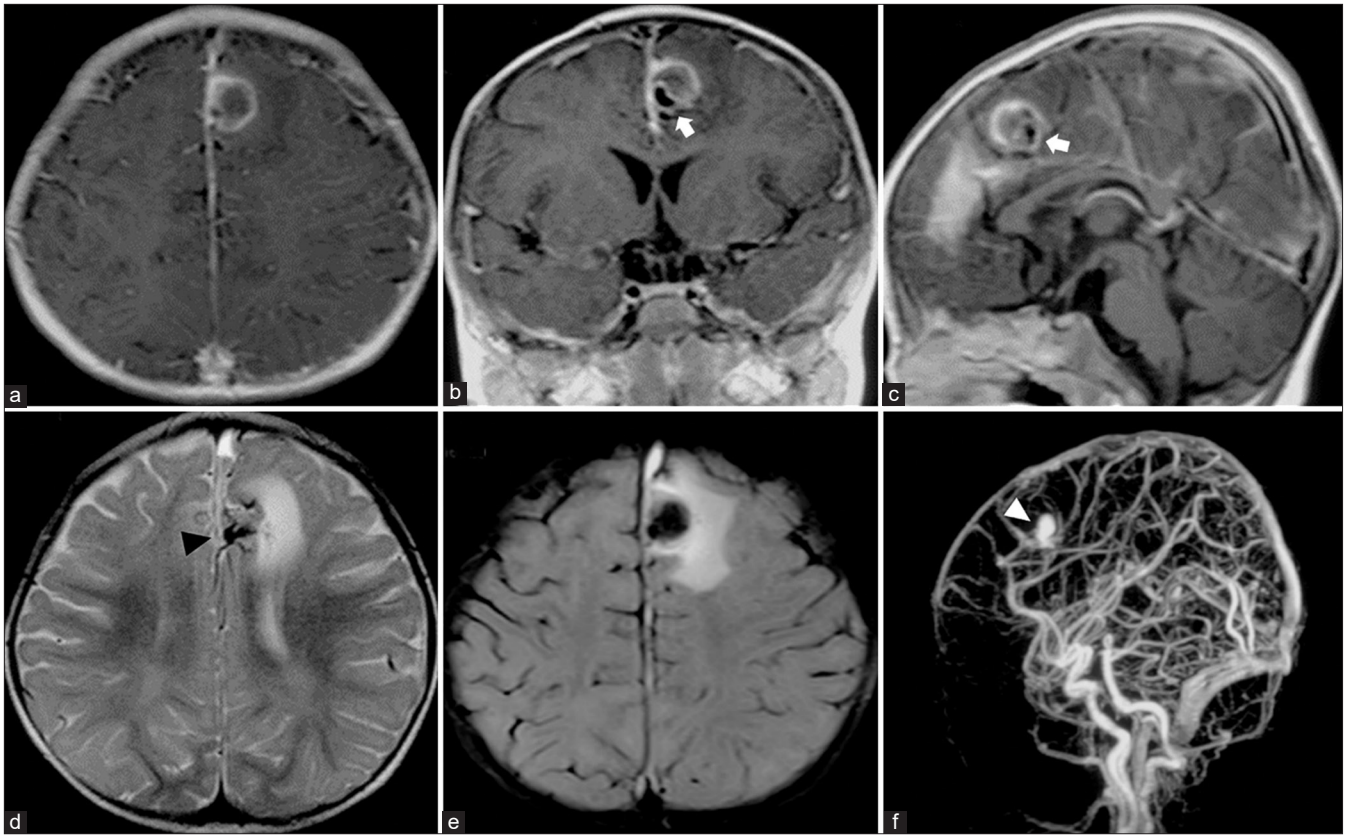

Figure 2: Magnetic resonance imaging (MRI) of the brain obtained 1 week after symptom onset. (a) Axial, (b) coronal, and (c) sagittal T1-weighted contrast-enhanced MRI show peripheral rim enhancing round mass with an eccentrically located signal void (white arrows), probably indicating a large partially thrombosed aneurysm. (d and e) Axial T2-weighted MRI reveals a tangle of abnormal flow voids (black arrowhead), probably representing pial arteriovenous malformations associated with a large aneurysm with the progression of perianeurysmal edema at the medial side of the left superior frontal region. (f) Sagittal maximum intensity projection image from 3D time-of-flight magnetic resonance angiography confirms the pial arteriovenous malformation with a large intranidal aneurysm (white arrowhead).

aneurysm, arising from the left middle internal frontal branches of the left anterior cerebral artery (ACA), classified as Grade I according to the Spetzler-Martin grading system. The AVM contained a 10 mm saccular aneurysm within the small nidus. The super selective injection with the microcatheter of the left ACA visualized a small nidus with multiple indirect small feeders, a large intranidal aneurysm, and early draining veins [Figure 4]. Due to the small indirect feeders, endovascular treatment was deemed unfeasible. Microsurgical resection was proposed but declined by her parents. A follow-up MRI 5 months later showed a significant reduction in the aneurysm size and complete resolution of the edema [Figure 5]. An electroencephalogram (EEG) indicated focal epileptic activity over the left parieto-occipital region, necessitating ongoing antiepileptic medication. Annual EEGs revealed similar patterns.

Six years after symptom onset, at age 7, a follow-up MRI and contrasted MRA confirmed the complete obliteration of the pial AVM and the intranidal aneurysm [Figure 6]. The patient exhibited normal development and had no recurrent seizures, achieving an average academic level.

At age 10, follow-up cerebral angiography disclosed a constellation of small, loosely packed vessels in the vicinity of the previously identified large thrombosed intranidal aneurysm. This area, devoid of early draining veins, was suggestive of focal hyperemia [Figure 7]. Progressing to age 12, advanced 3 Tesla MRI and contrast-enhanced MRA revealed a further diminution in the size of the aneurysm located at the medial part of the left superior frontal gyrus. In addition, an enhancing area was observed, corresponding to the prior angiographic findings of probable focal hyperemia [Figure 8].

By age 15, the patient had completed her middle school education but decided not to pursue further studies in high school. Around this time, she was diagnosed with depressive mood disorder and commenced treatment with sertraline

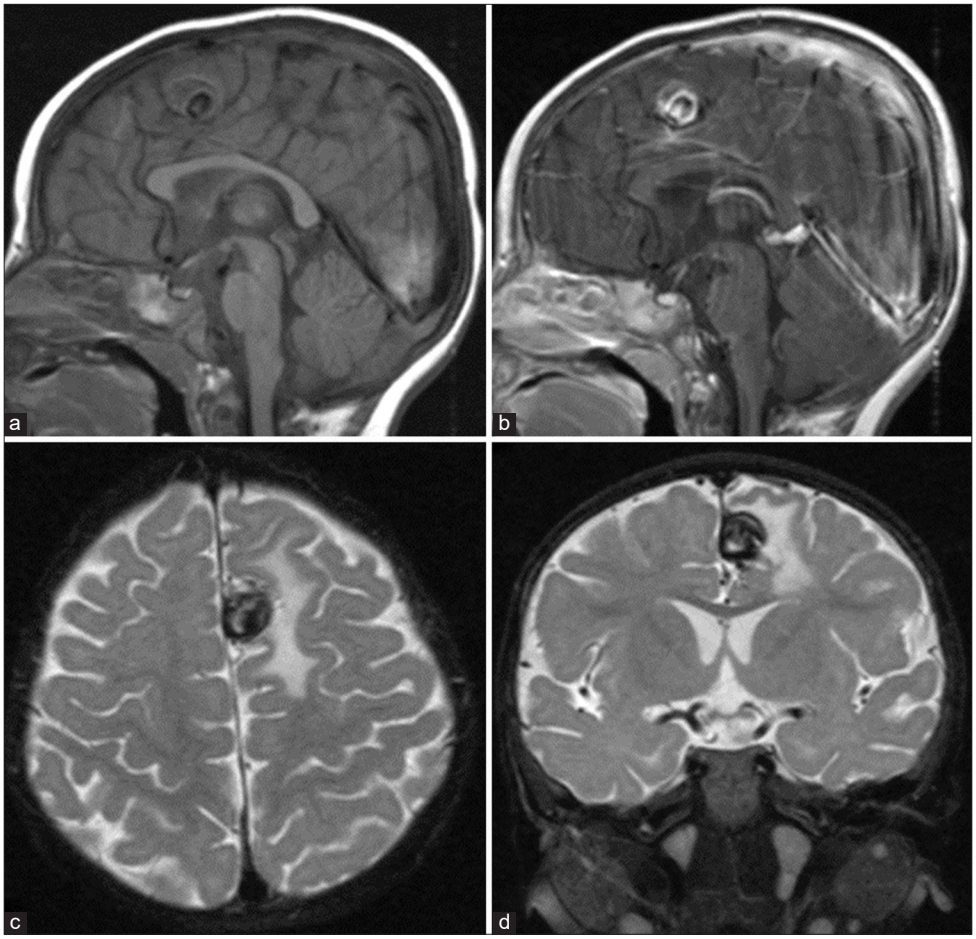

Figure 3: Magnetic resonance imaging (MRI) of the brain obtained 1 month after symptom onset. Sagittal T1-weighted MRI (a) with and (b) without contrast show heterogeneous signal intensity with eccentrically rim enhancing lesion, probably representing a laminated appearance of partially thrombosed aneurysm. (c) Axial and (d) coronal T2-weighted MRI reveals heterogeneous signal intensity in a large aneurysm surrounded by edema of the left superior frontal gyrus.

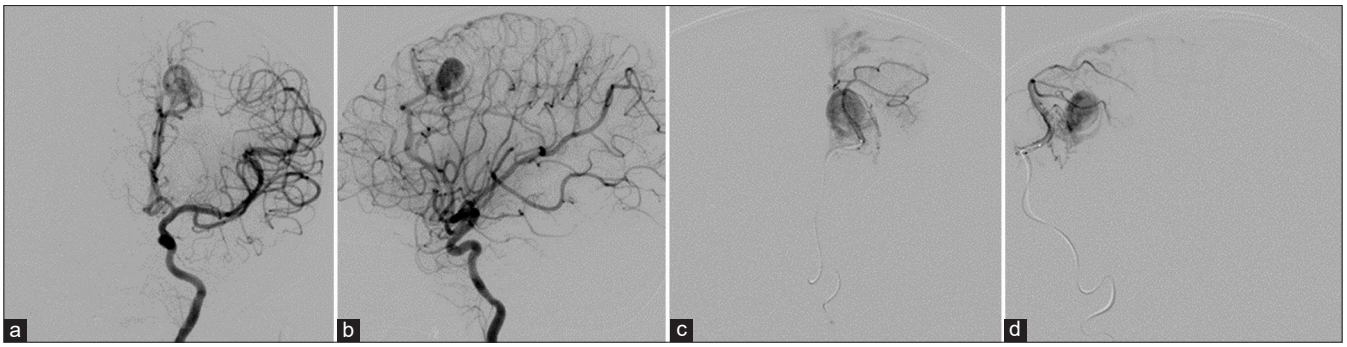

Figure 4: (a) Anteroposterior (AP) and (b) lateral views of the left internal carotid artery injection show a small pial arteriovenous malformation supplied by the left middle internal frontal branches of the left anterior cerebral artery (ACA) with early venous drainage into medial frontal veins and forward to superior sagittal sinus. A large aneurysm was located within the small nidus. (c) AP and (d) lateral views of the left ACA injection with microcatheter clearly demonstrate a small nidus, multiple indirect small feeders, large intranidal aneurysm, and early draining veins.

at a dose of 50 mg/day. At age 16, further imaging was undertaken, including a 3 Tesla cranial MRI and 4D dynamic contrast-enhanced MRA, which identified a recurrent pial AVM located in the left superior frontal region [Figure 9a-c].

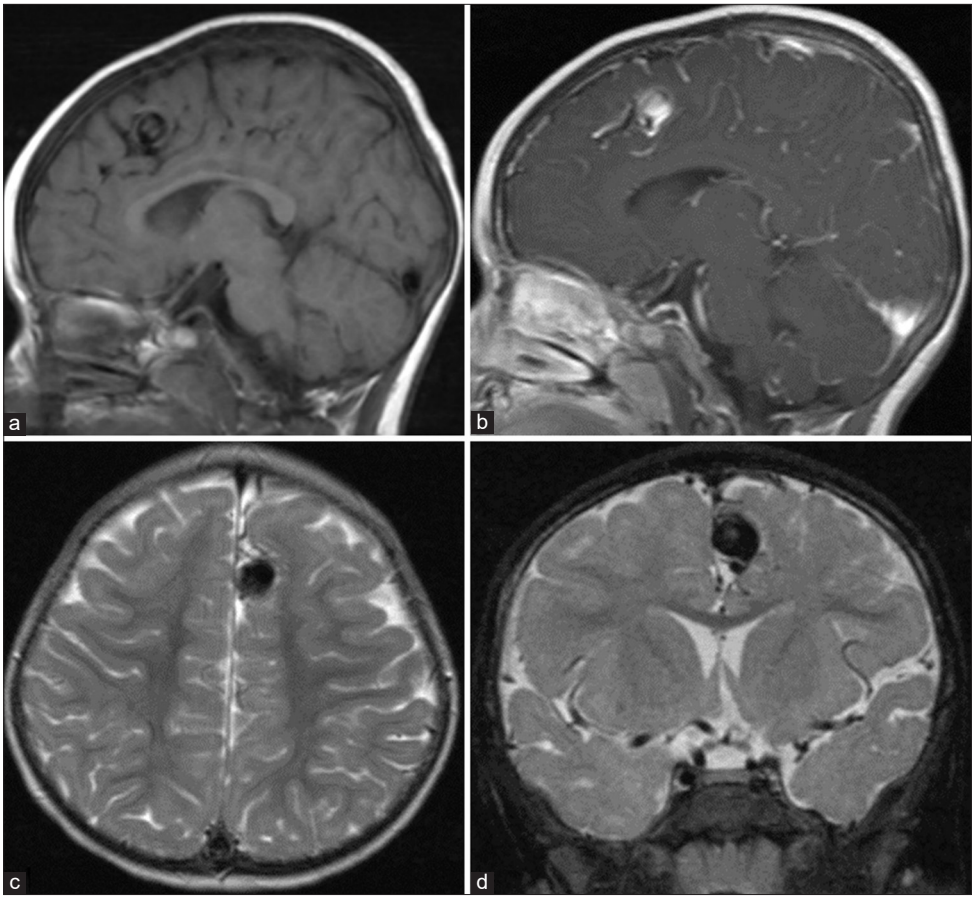

Figure 5: Magnetic resonance imaging (MRI) of the brain obtained 7 months after symptom onset. Sagittal T1-weighted MRI (a) with and (b) without contrast demonstrate a significant reduction in the size of a large partially thrombosed aneurysm. (c) Axial and (d) coronal T2-weighted MRI revealed a reduction in aneurysm size and complete resolution of perianeurysmal edema at the left superior frontal gyrus.

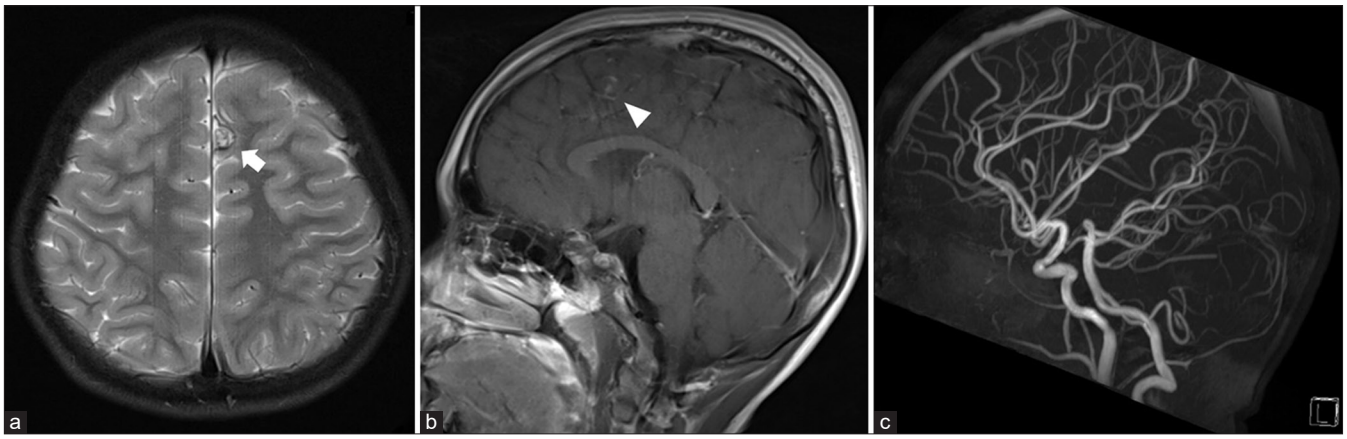

Figure 6: At age 7. (a) Axial T2-weighted magnetic resonance imaging (MRI) reveals a decrease in the size of the aneurysm (arrow) at the medial part of the left superior frontal gyrus. (b) sagittal T1-weighted contrast-enhanced MRI demonstrates complete thrombosis of the large aneurysm (arrowhead). (c) Sagittal maximum intensity projection image from 3D time-of-flight magnetic resonance angiography confirms complete obliteration of the pial arteriovenous malformation and large intranidal aneurysm.

Subsequent cerebral angiography performed 1 week later corroborated the findings of a recurrent pial AVM [Figure 9d and e]. Following a detailed discussion regarding treatment options with her parents, the decision was made to proceed with stereotactic radiosurgery for the recurrent AVM.

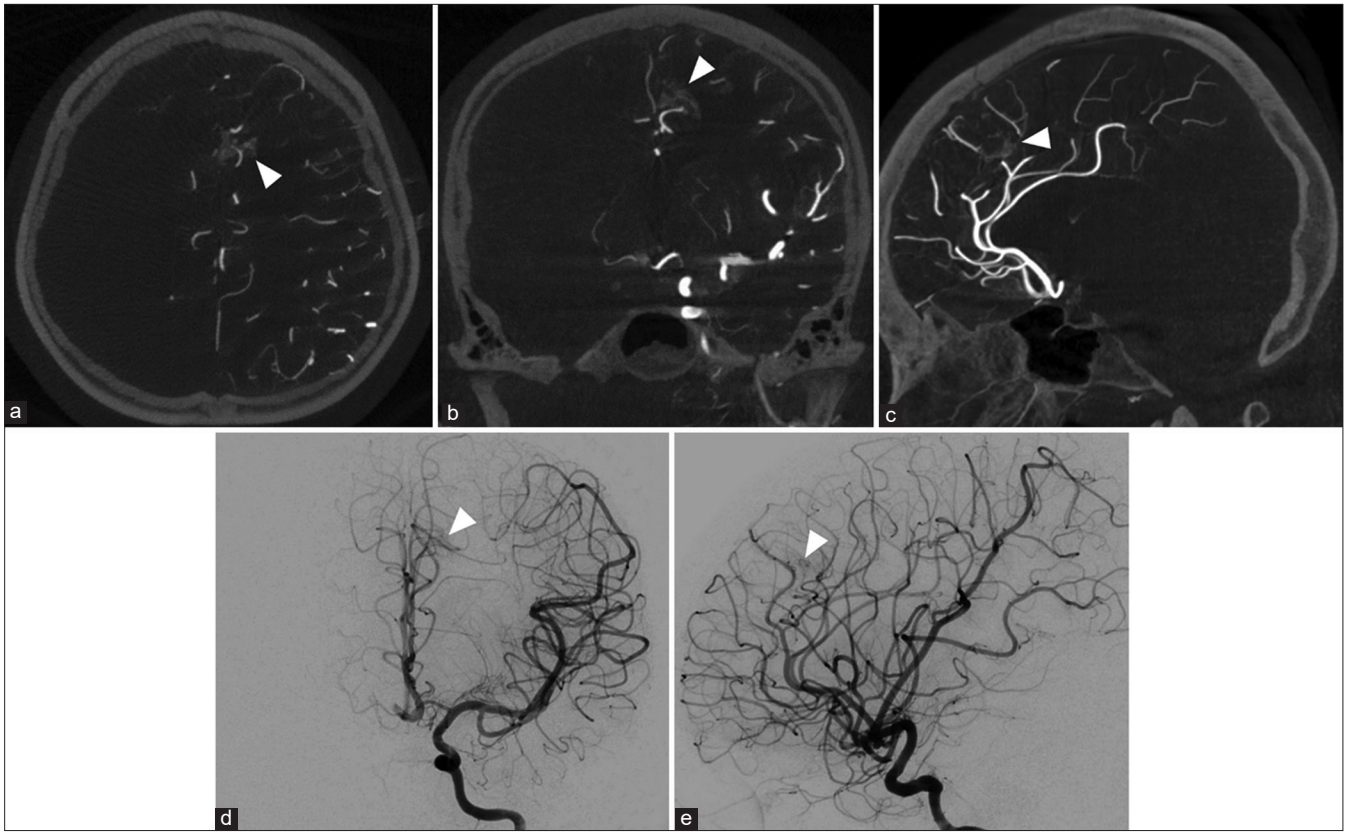

Figure 7: At age 10. (a) Axial, (b) coronal, and (c) sagittal views of maximum intensity projection reformatted image of angiographic computerized tomography, and (d) anteroposterior and (e) lateral views of the left internal carotid artery demonstrate a group of loosely packed tiny vessels (arrowheads) surrounding previous area of large thrombosed intranidal aneurysm without early draining vein, probably focal hyperemia. Arrowheads represent a group of loosely packed tiny vessels in all Figures 7a-7e.

DISCUSSION

Pediatric cerebral AVMs, primarily manifesting as hemorrhage in over 60% of cases, predominantly affect adolescents rather than younger children, with a rarity in infancy.[3,31] Based on the aged-related classification of pediatric cerebral AVMs, infant pial AVMs are distinct in their fistulous rather than glomerular architecture.[18] The pediatric ruptured brain AVMs carry an annual hemorrhage risk of 2–3% with deep venous drainage and associated aneurysms, heightening future bleeding risk.[4]

The primary treatment goal for pediatric ruptured brain AVMs is complete obliteration, preserving neurological function, and preventing further bleeding incidents.[3,8,31] Although microsurgical resection is often preferred, especially for low-grade AVMs (Spetzler-Martin I–II), endovascular treatment and radiosurgery are considered adjunctive. Intriguingly, children show a lower risk of subsequent bleeding compared to adults, prompting discussions around less aggressive treatment approaches.[9]

AVMs with associated aneurysms, found in 29% of pediatric cases, present a higher risk of rebleeding. The management of AVM-associated aneurysms, especially arterial ones, should not be delayed due to their strong correlation with hemorrhage.[3] Our patient's AVM, coupled with a large intranidal aneurysm, was deemed unsuitable for endovascular treatment, and surgical intervention was not pursued due to parental decision. Long-term imaging follow-up witnessed total thrombosis of the AVM and aneurysm.

Spontaneous obliteration of untreated cerebral AVMs is a rare phenomenon, and the incidence of this phenomenon was <1%.[1,5,20,22] Common factors in spontaneous thrombosis include a small nidus, hemorrhagic presentation, a single draining vein, and superficial venous drainage. The mass effect caused by parenchymal hemorrhage likely led to the stretching and narrowing of feeding arteries, the occlusion of draining veins, or the obliteration of the nidus, which contributed to the spontaneous obliteration of brain AVMs. The hematoma may have compressed the draining vein, causing partial or complete blockage of the vein. This blockage could result in venous stasis and the spread of a thrombus into the nidus of the AVM. Venous thrombosis in the main draining veins is likely a critical factor in the spontaneous regression of brain AVMs.[11,12] Our case

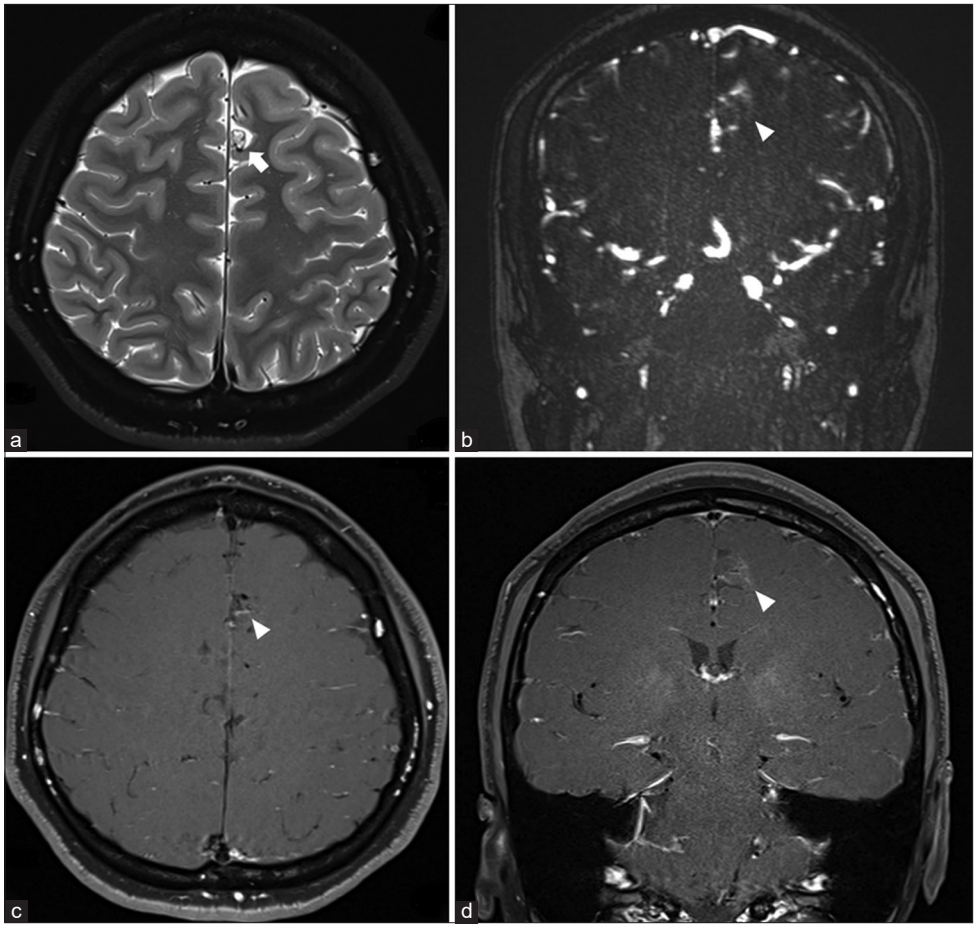

Figure 8: At age 12. (a) Axial T2-weighted magnetic resonance imaging (MRI) shows a further reduction in the size of the aneurysm (arrow) at the medial part of the left superior frontal gyrus. (b) Coronal view of contrast-enhanced magnetic resonance angiographic source image, and (c) axial and (d) coronal views of T1-weighted contrast-enhanced MRI reveal enhancing area (arrowheads) surrounding previous area of large thrombosed intranidal aneurysm, probably focal hyperemia, corresponding with prior cerebral angiography.

interestingly demonstrates thrombus formation beginning within the aneurysm progressing to complete thrombosis, with factors such as intracranial hemorrhage, small nidus, and lack of deep venous drainage being pivotal.

Spontaneous regression of pediatric AVMs is exceedingly rare, particularly in children under 5 years.[11,20] Since 1977, Dyck[7] reported spontaneous thrombosis of a brain AVM in a 4-year-old girl, who presented with a seizure. The surgery revealed a left parietal AVM supplied by distal branches of the ACA, featuring multiple distended and thrombosed venous channels but no evidence of hemorrhage. Histopathological examination confirmed extensive intravascular thrombosis. It was suggested that the thrombus propagation was due to elongation and tortuosity of the lesion caused by intravascular turbulence. In 1992, Iizuka *et al*.[15] reported on two pediatric cases of multiple cortical pial arteriovenous fistulas (AVFs) with significant spontaneous thrombosis. The first case involved a 6-month-old infant with four cortical AVFs, three of which underwent spontaneous thrombosis following a hemorrhage. The second case was a 1-year-old infant with seizures and two cortical AVFs, both showing spontaneous thrombosis, leading to total control of the AVFs. The causes of spontaneous thrombosis in these cases were not explored. In 2006, Leung *et al*.[21] reported that a case of spontaneous regression of cerebellar AVMs in a 4-year-old girl with hereditary hemorrhagic telangiectasia was reported. The AVM had a small nidus, a single draining vein, and no evidence of hemorrhage.

To the best of our knowledge, there was only one previous report of spontaneous disappearance of infant cerebral AVM by Mabe and Furuse.[23] They documented the spontaneous disappearance of a cerebral AVM in a 4-month-old infant over

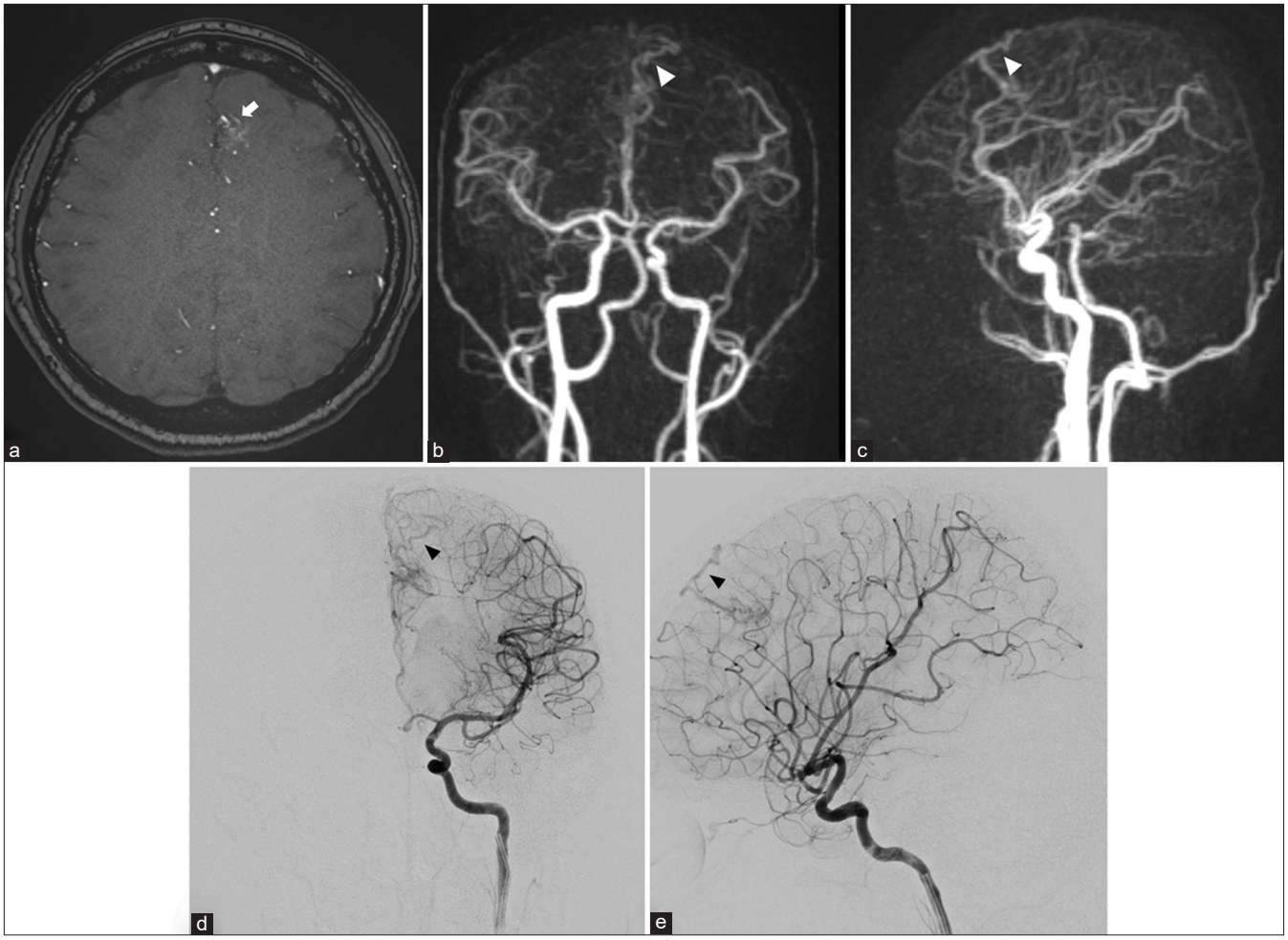

Figure 9: Imaging findings at 16 years of age. (a) Three-dimensional raw time-of-flight magnetic resonance angiography (MRA) source image demonstrates a cluster of diminutive vessels featuring an early draining vein (indicated by an arrow) located in the left superior frontal region. (b) Anteroposterior (AP) projection and (c) lateral projection of maximum-intensity 4D contrast-enhanced MRA reveal a recurrent pial arteriovenous malformation (AVM) characterized by an early draining vein (marked by white arrowheads). (d) AP and (e) lateral views of the left internal carotid artery injection confirm the presence of a recurrent pial AVM with an early draining vein (denoted by black arrowheads).

7 months. The infant presented with impaired development and increased head circumference due to hydrocephalus. A postoperative SDH following a ventriculoperitoneal shunt might have been a contributing factor.

At present, two cases of spontaneous resolution of cerebral pial AVFs in children have been reported. The first case, described by Sabrina *et al*.,[29], involved an asymptomatic preterm newborn who was incidentally found to have a pial AVF, which spontaneously resolved within 14 days of life without any intervention. The reasons for this phenomenon remain unclear. The second case, reported by Chen *et al*.,[6], involved a 2-month-old boy presented with seizures and multifocal subpial hemorrhage. He underwent transarterial embolization, and at an 8-month follow-up, additional pial fistulas were discovered but had spontaneously thrombosed 2 weeks later. The speculated cause was vessel kinking during vascular remodeling, leading to auto-stenosis, flow restriction, and subsequent thrombosis.

Interestingly, persistent focal hyperemia was observed during follow-up in our case, involving a patient aged 10–12, through control angiography and serial MRI/MRA examinations. The underlying pathophysiology of focal hyperemia, however, remains not fully understood. Neuronal activity in the brain triggers localized increases in blood flow, a phenomenon known as functional hyperemia. Essentially, when there is a surge in brain activity in a particular area, blood flow to that specific region also increases. This ensures that active neurons receive the necessary nutrients. Functional hyperemia refers to this elevation in local blood flow, leading to the dilation of blood vessels, as a direct response to neuronal activity.[26]

Focal hyperemia often follows traumatic brain injury, particularly in patients with focal contusions and intracerebral

hematomas, affecting both gray and white matter. This hyperemia typically arises in the healthy tissue directly adjacent to focal mass lesions. During the healing process, neovascularization occurs around the contused brain area. However, these new blood vessels lack tight endothelial junctions, leading to disturbances in the blood-brain barrier. This type of focal and persistent hyperemia is considered a benign form of hyperemia. It generally has a minimal impact on intracranial pressure and the patient's level of consciousness and is associated with a better patient outcome.[10,30] However, focal hyperemia could be an early indicator of the recanalization of a cerebral AVM.[19] Therefore, it is essential to conduct further long-term follow-up imaging studies, especially angiography, in our case.

Recurrent cerebral AVMs after angiographically documented complete excision may develop particularly in children, highlighting the importance of long-term imaging follow-up. Even though the likelihood of recurrence following a negative postoperative angiogram is low, the associated morbidity and mortality due to hemorrhage are significant.[2,17,25,28] In addition, there is the possibility of recanalization of spontaneously obliterated pial AVMs.[24,27] Despite a low probability of recanalization, the actual prevalence might be underreported due to insufficient follow-up after obliteration.[22] To evaluate the durability of spontaneous obliteration, long-term monitoring through clinical follow-up and serial imaging is crucial, particularly with delayed control angiography. While follow-up angiography remains the gold standard for confirming cerebral AVM, it has drawbacks such as exposure to ionizing radiation, invasiveness, the frequent need for general anesthesia, preparation requirements, and costs.[9,29] MRI and contrast-enhanced MRA are fast, noninvasive alternatives that can confirm the stability of a complete occlusion. However, some experts caution against relying solely on MRI/MRA for assessing occlusion stability.[2,19] With advancements in neuroimaging, high-resolution dynamic MRA techniques, such as 4D MRA, may become routine in pediatric AVM follow-up, offering excellent angiographic quality.[16,32]

In the present case, the transition from focal hyperemia to the recurrence of a pial AVM was observed, illustrating a rare and clinically significant progression. Initially, focal hyperemia was noted in the regions surrounding the previously obliterated AVM. Typically, focal hyperemia indicates increased blood flow that can occur in response to localized neuronal activation or as a reactive process following vascular injury. In this case, it was hypothesized to be an adaptive response to the altered hemodynamics postinitial AVM obliteration. Over time, this area of hyperemia began to show signs of neovascularization. Neovascularization following AVM treatment, particularly in cases involving radiosurgery or surgical interventions, is well-documented, but its progression to AVM recurrence is not well understood and rarely documented. The development of a recurrent AVM from focal hyperemia in our patient underscores a critical aspect of pediatric AVM management: it particularly highlights the potential for angiogenic stimuli within the brain to promote the formation of abnormal vascular networks. Advanced imaging techniques, including dynamic contrast-enhanced MRA, were instrumental in identifying and monitoring these changes. These imaging findings were pivotal in distinguishing between benign hyperemia, which typically resolves without clinical sequelae, and the more concerning pattern suggestive of neovascular activity leading to AVM recurrence.

CONCLUSION

We reported an exceptionally rare case of spontaneous regression in a pial AVM concurrent with a large intranidal aneurysm. Extensive long-term follow-up and sequential imaging have meticulously documented the evolution and eventual complete thrombosis of both the substantial partially thrombosed aneurysm and the smaller pial AVM. This spontaneous regression was notably influenced by factors such as a hemorrhagic presentation, superficial venous drainage, and a small nidus. In addition, our observations confirmed the complete resolution of perianeurysmal edema.

It is crucial to recognize that spontaneous thrombosis of ruptured cerebral AVMs remains a rare and unpredictable phenomenon. Therefore, it should not be considered a reliable treatment strategy in the management of brain AVMs. Instead, meticulous long-term clinical and radiological monitoring is imperative for pediatric patients who experience spontaneous obliteration of cerebral AVMs.

Moreover, our case provides a significant example of focal hyperemia developing into a recurrent pial AVM, illustrating a dynamic and critical area of pediatric neurovascular care. This progression prompts the need for further research and could potentially impact future treatment approaches and monitoring protocols in pediatric neurovascular disorders.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

Patients' consent not required as patients' identities were not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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How to cite this article: Iampreechakul P, Wangtanaphat K, Wattanasen Y, Hangsapruek S, Lertbutsayanukul P, Siriwimonmas S. Long-term surveillance in an infant with spontaneous obliteration of pial arteriovenous malformation and large intranidal aneurysm: A unique case observation. Surg Neurol Int. 2024;15:206. doi: 10.25259/SNI_45_2024