# Development of a de novo arteriovenous malformation after severe traumatic brain injury

Case report

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Arteriovenous malformations (AVMs) are typically considered congenital lesions, although there is growing evidence for de novo formation of these lesions as well. The authors present the case of an AVM in the same cerebral cortex that had been affected by a severe traumatic brain injury (TBI) more than 6 years earlier. To the best of the authors' knowledge, this is the first report attributing the formation of an AVM directly to TBI. (*http://thejns.org/doi/abs/10.3171/2014.7.PEDS1431*)

KEY WORDS • AVM • arteriovenous malformation • traumatic brain injury • vascular disorders • trauma

A RTERIOVENOUS malformations (AVMs) are generally considered to be congenital lesions that are discovered either incidentally or after becoming symptomatic, most often due to hemorrhage. Several case reports have described the de novo development of AVMs when prior brain imaging has been performed.<sup>1,4,19</sup> We present the unique case of a child who had sustained a severe traumatic brain injury (TBI), underwent decompressive hemicraniectomy and cranioplasty, and later developed a de novo AVM in the injured cortex.

# **Case Report**

*History and Examination*. An 11-month-old girl with no remarkable medical history presented to an outside hospital after falling from her parents' porch. Initial head CT demonstrated left-sided skull fractures and a subdural hematoma. The patient was immediately taken for clot evacuation and decompressive hemicraniectomy and was then transferred to our pediatric hospital. She underwent cranioplasty 10 days after her first surgery and made a good recovery. At 22 months of age, she underwent revision cranioplasty at our institution for a persistent skull defect. She had recovered well from her injury with intact speech and the ability to ambulate without assistance, albeit with persistent right hemiparesis. At 6 years of age, she developed a seizure disorder. Magnetic resonance imaging of her brain was performed at that time and demonstrated encephalomalacia at the site of the prior injury with no evidence of a vascular malformation (Fig. 1 left). Her seizures were well controlled with medication until she was 12 years old, when her seizure frequency increased to approximately two episodes per week. In response to this change, she underwent repeat MRI, which revealed a vascular malformation within the large area of encephalomalacia in the left parietal lobe (Fig. 1 right). Angiography demonstrated an AVM measuring 2.5 cm in diameter, fed predominantly from the left pericallosal artery and draining into the superior sagittal sinus (Fig. 2). The patient had no clinical signs or radiological evidence of hemorrhage from the AVM.

*Operation*. Shortly after angiography, the patient underwent elective resection of the AVM. The feeding arteries and then the draining veins were coagulated, and the AVM was removed en bloc. Intraoperative visualization and pathological examination of tissue revealed a typical AVM with no unusual characteristics (Fig. 3).

*Postoperative Course*. There were no surgical complications, and the patient made an uneventful recovery. Three weeks after surgery, she returned to the clinic at her neurological baseline. Although she experienced a cluster

*Abbreviations used in this paper:* AVM = arteriovenous malformation; TBI = traumatic brain injury.

This article contains some figures that are displayed in color online but in black-and-white in the print edition.

# Posttraumatic arteriovenous malformation

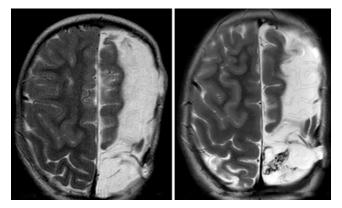


Fig. 1. Axial T2-weighted MRI studies obtained before and after development of an AVM. Left: Image obtained when the patient presented with seizures at 6 years of age, demonstrating encephalomalacia and no vascular lesion. Right: Image obtained when the patient was 12 years of age, demonstrating left parietal vascular malformation.

of seizures on postoperative Day 1, she has been seizure free for 2 months since then.

## Discussion

This is the first reported case of an AVM developing within traumatized brain. Arteriovenous malformations are generally considered to be congenital lesions, which are discovered either incidentally or after hemorrhage. De novo AVMs have been described in both children and adults,<sup>4</sup> with the majority of lesions being noted in younger, female patients<sup>13</sup> similar to our patient. There are case reports of de novo AVM formation in children associated with cavernous malformations<sup>2</sup> or moyamoya disease<sup>15,18</sup> or without any previous vascular pathology.<sup>13,19</sup> In a study of younger patients with AVMs, almost 10% of the patients had undergone previous angiography without evidence of vascular malformation.11 There are also multiple case reports of AVMs presenting with subdural hemorrhage,<sup>5,17</sup> but no reports that we are aware of attribute an acute supratentorial subdural hemorrhage to an AVM.

The occurrence of other vascular pathologies, such as aneurysms and dural arteriovenous fistulas, following head trauma have been well documented.<sup>3,16</sup> To our knowledge, only one prior report has described the development of an AVM in a child after head trauma, and that

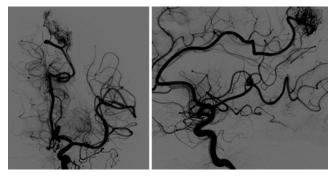


Fig. 2. Axial (left) and sagittal (right) angiograms obtained after a second MRI study, demonstrating a left parietal AVM fed from branches of the pericallosal artery.

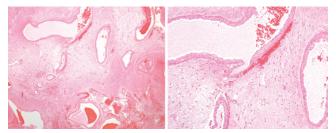


Fig. 3. Photomicrographs of biopsied AVM tissue demonstrating the typical morphology of tortuous pathological vessels. H & E, original magnification ×40 (left) and ×100 (right).

case is substantially different from the one we present here.<sup>7</sup> The AVM in that patient developed within radiographically normal brain tissue distant from the site of the injury. Furthermore, that patient had suffered a less severe TBI, with only a small amount of traumatic subarachnoid hemorrhage. Therefore, the authors believed that the AVM could not be definitively related to the prior head injury.

The growing number of reports on de novo AVMs together with reports on the growth and regression of AVMs<sup>6</sup> is evidence that these lesions are dynamic, rather than static, entities. There is a small possibility that our patient harbored an AVM from the outset of her injury and that it went undetected until her second MRI study. However, we do not believe this to be the case, as MRI has been shown to be 97% sensitive in detecting unruptured AVMs.9 Our patient's first MRI study was of good quality and was reviewed by neurosurgeons and a pediatric neuroradiologist with knowledge of the subsequent MRI findings. Furthermore, our patient's medical history was clear. Her parents described a fall and resultant head trauma without any antecedent neurological changes, and there were no findings on her initial posttraumatic imaging that suggested anything other than typical subdural hemorrhage and skull fracture. Neuroimaging in the subacute and chronic phase of TBI has demonstrated hippocampal atrophy, white matter connectivity changes, and defined areas of encephalomalacia that may be resected for the treatment of epilepsy.8,10,20 While there are no specific guidelines on repeat imaging in patients during the subacute or chronic phase of TBI, the present case emphasizes the fact that new or worsening symptoms remote from a TBI still warrant radiographic investigation.

## Conclusions

In summary, to our knowledge, this is the first report documenting the development of an AVM in response to severe TBI. There is a theory that de novo AVM development can be attributed to a "two-hit" phenomenon in which trauma may serve as the "second hit."<sup>2</sup> Our patient had no medical or family medical history to suggest a first hit, leading us to ascribe the development of this AVM solely to the severe TBI she had suffered years before the AVM was discovered. Much research has shown that angiogenic factors, such as vascular endothelial growth factor and stromal cell–derived factor 1, increase in the days following a TBI,<sup>12,14</sup> but the subacute release of these factors is unlikely to account for the protracted development of the AVM a decade later. Thus, the underlying mechanism linking the injury to the AVM remains unclear.

#### Disclosure

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

Author contributions to the study and manuscript preparation include the following. Conception and design: Miller, Chern. Acquisition of data: all authors. Analysis and interpretation of data: all authors. Drafting the article: Miller, Bass. Critically revising the article: all authors. Reviewed submitted version of manuscript: Miller.

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