



## Review

## Arteriovenous malformations and headache



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## ABSTRACT

Brain arteriovenous malformations (AVM) are complex vascular lesions commonly associated with chronic headache. An occipital location appears to increase the risk of concurrent migraine-like headaches in AVM patients. We have experienced great success in treating these headaches through a multidisciplinary approach to eradicate cerebral AVM. However, the specific clinical characteristics of AVM-associated headaches and the most effective treatment strategies for these patients remain unclear. Here, we provide a comprehensive review of the literature on AVM-associated headaches. We detail the history, classification, epidemiology, presentation, pathophysiology, treatment options, and outcomes for this poorly described condition. Additionally, we illustrate our approach to the management of patients with occipital AVM and associated intractable headaches.

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## 1. Introduction

Arteriovenous malformations (AVM) of the brain are complex tangles of abnormal vessels that result in the direct arteriovenous shunting of blood due to lack of an intervening capillary bed. Although hemorrhage is the most common reason for initial presentation, a significant subset of patients may present with debilitating headaches that are not due to hemorrhage [1]. The International Classification of Headache Disorders (ICHD-3) outlines the diagnostic criteria for headaches attributable to AVM (Table 1) [2]. According to ICHD-3 criteria, AVM-associated headaches have a clinical course that parallels that of a coexisting AVM in terms of temporality, severity, and location.

Hoffman first diagnosed intracranial AVM clinically in 1889, and the initial report of migraine-like headache associated with an AVM came from De Lange in 1927 [3–5]. Early studies further explored the existence of AVM-associated migraine “sequences” [6,7]. Although some believe that AVM-headache syndromes are clinically indistinguishable from migraine without AVM, the association between occipital AVM and headache is well recognized [8]. Prominent visual and other migraine-like symptoms are associated with occipital AVM [5,9–19].

The basis for an association between AVM and headache remains speculative and is not well understood. Clinically distinguishing headaches that are associated with an AVM from those

that are not is also challenging [8,20,21]. Thus, we have reviewed the literature on AVM-associated headaches to consolidate information on this challenging clinical entity. The comprehensive multidisciplinary approach we utilize in the management of patients with unruptured AVM and associated intractable headaches is also showcased.

## 2. Case illustration

## 2.1. Presentation

A 35-year-old woman with a lifelong history of migraine-like headaches presented for evaluation. Her headaches were stereotypical, occurred only on the right side and were preceded by visual auras and photophobia. Due to increasing headache frequency a MRI was obtained, revealing a 3 cm right parieto-occipital AVM (Fig. 1). Catheter cerebral angiography confirmed the presence of a Spetzler–Martin Grade 3 AVM fed by branches of the middle cerebral, posterior cerebral, and middle meningeal arteries. Given the constellation of clinically and anatomically concordant findings, this patient’s headaches were determined to be AVM-related. A treatment plan with a goal of AVM eradication was elected.

## 2.2. Intervention

Preoperative endovascular embolization of the AVM was performed with the liquid embolic agent n-butyl cyanoacrylate on

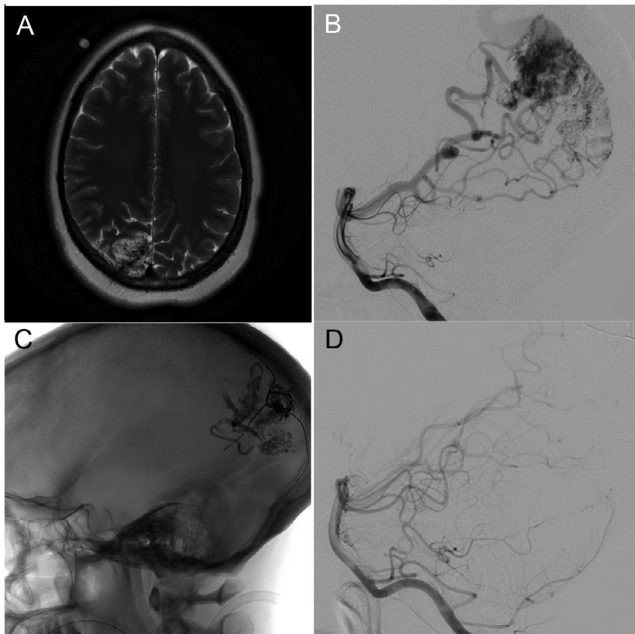
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**Table 1**  
Criteria for arteriovenous malformation-associated headache\*

- (A) An AVM has been diagnosed
- (B) Evidence exists of causation where:
  1. Headache led to the discovery of the AVM or developed in close temporal relation to AVM signs and symptoms
  2. Headache improved/worsened concurrently with the clinical course of the AVM
- (C) Headache is localized to the site of the AVM
- (D) Headache is not better accounted for by another headache disorder and intracranial hemorrhage has been excluded

\*Adapted from *International Classification of Headache Disorders (ICHD-3)*.  
AVM = arteriovenous malformation.



**Fig. 1.** Occipital arteriovenous malformation (AVM) in a patient with headaches. Preoperative T2-weighted axial MRI demonstrates flow voids in the right parieto-occipital region consistent with a brain AVM (A). Left vertebral arteriogram in lateral projection confirms a 3 cm AVM arising from distal branches of the right posterior cerebral artery (B). Later skull radiograph shows radio-opaque glue cast after preoperative n-butyl cyanoacrylate (NBCA) embolization (C). Postoperative catheter angiography after vertebral artery injection confirms that a complete resection of the AVM was accomplished (D).

the day prior to scheduled AVM resection. Targeted embolization was performed with the primary goal of eliminating deep arterial feeders and secondarily to reduce global AVM blood flow. The patient tolerated embolization well although a left homonymous hemianopia with macular sparing was detected. On the following day the patient underwent uneventful craniotomy for complete AVM resection (Fig. 1). Immediate postoperative catheter angiography demonstrated no further arteriovenous shunting.

### 2.3. Postoperative course

Postoperatively, the patient noticed a reduction in the frequency and intensity of her prior headaches. She was discharged home on the third postoperative day. At follow-up 2 months later she reported that she was completely headache-free.

Review of the senior author's (R.A.S.) database of unruptured occipital AVM patients who underwent resection revealed the case result to be generalizable. Of 16 such patients who presented with intractable headaches or migraine between 1998 and 2013, all reported significant improvement or resolution of headache after complete AVM resection.

## 3. History and classification

The diagnosis of brain AVM was classically made after exploratory craniotomy in a patient presenting with focal seizure. Both Cushing and Dandy published series of brain “angiomas” in the 1920s in patients presenting this way [22,23]. Cushing and Bailey, however, were among the first to demonstrate the connection between brain AVM and headaches. In their series of 14 brain “angiomas” published in 1928, three patients were noted to have occipital AVM presenting with headache as the main complaint [6,23].

In the years that followed, the association between AVM and headache became increasingly reported in the literature. In 1940, Northfield described a patient with 37 years of headache with a migraine-like quality that occurred on the side of his AVM [24]. Olivecrona and Riives (1948) published their experience with 42 patients, noting that headache was common and that each patient with headache had an occipital AVM [25]. In their series, however, only one patient had symptomatology consistent with typical migraine-headache preceded by scintillating scotomas. Mackenzie (1953) found that 12 (24%) of 50 consecutive patients presented with headache as their initial AVM symptom, seven of which were “suggestive of migraine” [6].

The ICHD-3 indicates that AVM can cause attacks of migraine [2]. It is noted that up to 58% of women with AVM have migraine with aura as the presenting symptom. A strong correlation between the hemispheric side of an AVM and the side of headache or aura is thought suggestive. Evidence linking AVM to other headache disorders such as cluster headache, paroxysmal hemicrania, and neuralgiform headache is less robust.

## 4. Epidemiology and presentation

AVM have a prevalence of approximately 0.1% in the population [26]. The incidence of AVM-associated headache is unknown. In one study, only 0.2% of patients with a normal neurological examination who underwent neuroimaging for headache had an AVM [27].

Localization of headache to the side of the AVM is common but not mandatory [10,12]. Occipital AVM location is thought to be a risk factor for headaches [9,10,12,28]. These patients typically have concurrent visual symptoms including field cuts, blurring, scintillations, and/or diplopia [12].

### 4.1. Headache

A wide range of values for the frequency of headache as a presenting symptom of AVM has been reported in the literature. Hofmeister et al. found that 14% of 1,289 AVM patients reported chronic headaches [1]. Crawford et al. reported a less robust association with headache in only three of 343 AVM patients at the time of diagnosis [29]. In a small series of 48 AVM patients, Wal-timo reported that 79% had headache disorders [28]. Hartmann

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