



An aggressive dural arteriovenous fistula manifested by unilateral subcortical calcification and cerebral edema: A case report

Kagawa, Yuya ; Imahori, Taichirou ; Okino, Reiichi ; Harada, Tomoaki ;
Yamamoto, Daisuke ; Miyake, Shigeru ; Sasayama, Takashi

(Citation)

Radiology Case Reports, 18(11):4218-4221

(Issue Date)

2023-11

(Resource Type)

journal article

(Version)

Version of Record

(Rights)

© 2023 The Authors. Published by Elsevier Inc. on behalf of University of Washington.
This is an open access article under the Creative Commons Attribution-NonCommercial-
NoDerivatives 4.0 International license

(URL)

<https://hdl.handle.net/20.500.14094/0100483375>



Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.elsevier.com/locate/radcr

Case Report

An aggressive dural arteriovenous fistula manifested by unilateral subcortical calcification and cerebral edema: A case report ☆☆☆

Yuya Kagawa, MD^a, Taichiro Imahori, MD, PhD^{a,b,*}, Reiichi Okino, MD^a,
Tomoaki Harada, MD, PhD^a, Daisuke Yamamoto, MD, PhD^a, Shigeru Miyake, MD, PhD^a,
Takashi Sasayama, MD, PhD^b

^a Department of Neurosurgery, Kitaharima Medical Center, Hyogo, Japan

^b Department of Neurosurgery, Kobe University Graduate School of Medicine, Hyogo, Japan

ARTICLE INFO

Article history:

Received 31 July 2023

Revised 19 August 2023

Accepted 21 August 2023

Available online 19 September 2023

Keywords:

Calcification

Brain edema

Central nervous system vascular malformations

Radiology

Interventional

Brain Neoplasms

ABSTRACT

Unilateral subcortical calcifications are unique radiographic findings indicating specific focal pathologies. When the lesion is accompanied by edema, cerebral neoplasm usually leads to a differential diagnosis. This report presents a case of unilateral subcortical calcification and edema that resulted in cerebral hemorrhage and a subsequent diagnosis of an aggressive dural arteriovenous fistula. A man in his 60s presented with left hemianopsia and a progressive headache for over 6 months. Initial computed tomography revealed unilateral subcortical calcification and cerebral edema in the right occipital lobe, raising the suspicion of oligodendroglioma. However, 10 days later, a cerebral hemorrhage occurred in the lesion. Magnetic resonance imaging revealed flow void clusters and dilatation of the bilateral external carotid arteries and cortical veins, indicating a dural arteriovenous fistula. Cerebral angiography confirmed the presence of a parasagittal dural arteriovenous fistula (Borden type III). The patient was successfully treated with trans-arterial embolization using Onyx. Thus, calcifications with edema are more commonly associated with cerebral neoplasms; however, in this case, they indicated the presence of a dural arteriovenous fistula with severe corticovenous reflux. The presented case highlights the importance of recognizing these imaging features in dural arteriovenous fistulas and raises awareness of the potential danger of early hemorrhage after diagnosis. Therefore, timely evaluation of cranial vessels is essential in cases of unilateral subcortical calcification and edema to facilitate the early detection and management of aggressive dural arteriovenous fistulas.

© 2023 The Authors. Published by Elsevier Inc. on behalf of University of Washington.

This is an open access article under the CC BY-NC-ND license
(<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

☆ Acknowledgments: Part of this research was supported by the Japan Society for the Promotion of Science Grants-in-Aid for Scientific Research (JSPS KAKENHI grant numbers 21KK0289 and 23K15668).

☆☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

* Corresponding author.

E-mail address: taichiro.imahori@gmail.com (T. Imahori).

<https://doi.org/10.1016/j.radcr.2023.08.097>

1930-0433/© 2023 The Authors. Published by Elsevier Inc. on behalf of University of Washington. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Introduction

Unilateral subcortical calcifications provide valuable radiographic insights into the clinical diagnosis because they are associated with specific focal pathologies [1,2]. These calcifications are occasionally identified in cerebral neoplasms, with a particularly high prevalence in oligodendrogliomas [3]. Moreover, this calcification type is uncommon yet occasionally found in vascular disorders involving arteriovenous shunts, such as dural arteriovenous fistulas (DAVFs) [4–6]. These radiographic findings indicate a chronic clinical course because calcification develops over a relatively long period [6]. However, some arteriovenous shunt diseases progress rapidly to the clinical stage with severe corticovenous reflux (CVR), requiring immediate management [7–9]. Therefore, for timely therapeutic intervention, in addition to calcifications, other potential radiographic features should be identified and recognized.

In this report, we present a case of unilateral subcortical calcification concomitant with cerebral edema, which subsequently developed into cerebral hemorrhage and was treated for an aggressive DAVF.

Case presentation

A man in his 60s presented with left hemianopsia and headache that had gradually progressed over the past 6 months. The patient had a history of traumatic subarachnoid hemorrhage treated conservatively and leukemia cured with chemotherapy 13 and 8 years prior, respectively, but no current medications for other diseases. Computed tomography (CT) performed at the outpatient clinic revealed unilateral subcortical calcification concomitant with cerebral edema in the right occipital lobe (Fig. 1). Based on clinical and imaging findings, oligodendroglioma was initially considered the most probable diagnosis. Further examinations at the outpatient clinic and an elective open resection surgery were scheduled.

However, 10 days later, the patient suffered a sudden left hemiparalysis, and a cerebral hemorrhage anterior to the right occipital lesion was confirmed via CT (Fig. 2). Magnetic resonance imaging showed flow void clusters within the lesion, while magnetic resonance angiography revealed dilatation of the bilateral external carotid arteries and cortical veins of the right occipital lobe, indicating the presence of a DAVF (Figs. 3A–C).

Cerebral angiography revealed a marked CVR around the right occipital lobe from the shunt at the parasagittal sinus, with the bilateral middle meningeal and occipital arteries as the main feeding arteries, confirming a diagnosis of parasagittal DAVF (Borden type III) (Fig. 4A). Four days after the hemorrhagic presentation, the shunt was successfully obliterated via trans-arterial embolization using Onyx (Medtronic, Minneapolis, MN) (Figs. 4B and C).

After treatment, the left hemiparalysis improved, with a reduction in the edema on CT. The patient was then transferred

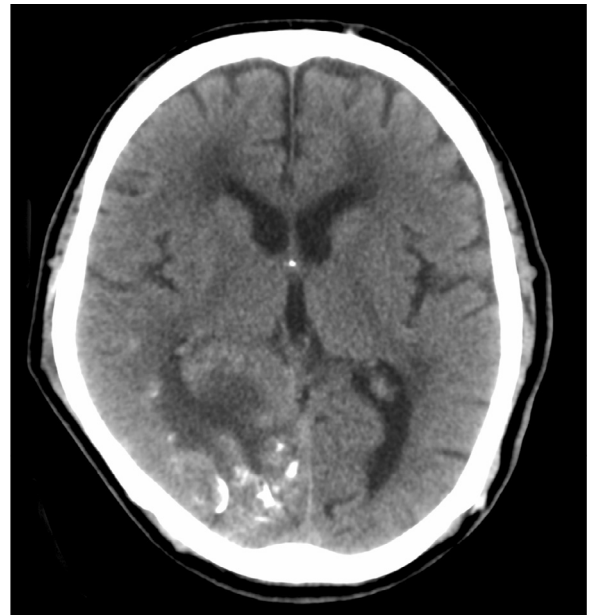


Fig. 1 – Computed tomography at the time of the initial visit. Nonenhanced computed tomography showing multiple curvilinear calcifications with cerebral edema in the subcortical region of the right occipital lobe.



Fig. 2 – Computed tomography 10 days after the initial visit. Nonenhanced computed tomography showing hemorrhage in the anterior part of the right occipital lobe (arrow).

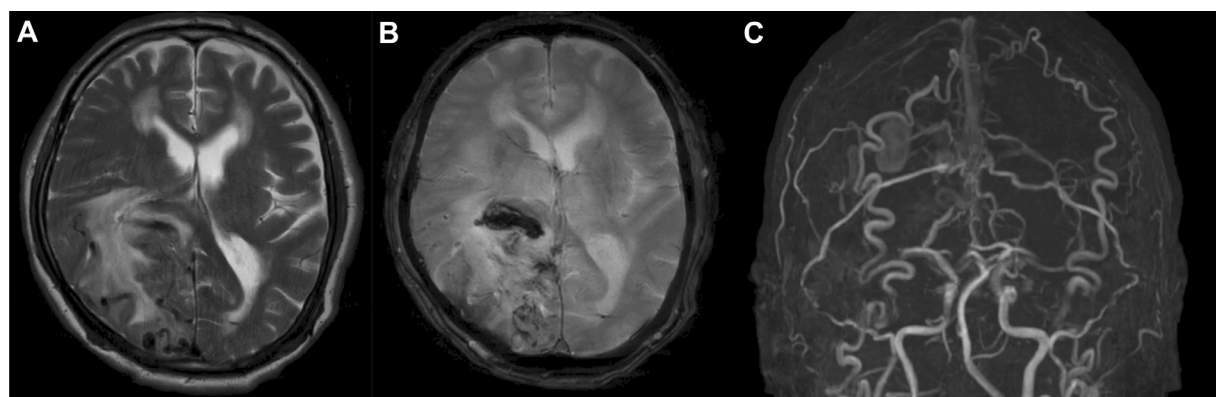


Fig. 3 – Magnetic resonance imaging after the occurrence of the hemorrhage. (A) T2-weighted image showing flow-void clusters in the right occipital lobe. (B) T2*-weighted image showing hemorrhage in the anterior part of the right occipital lesion. (C) Time of flight magnetic resonance angiography showing dilatation of the bilateral external carotid arteries and cortical veins of the right occipital lobe.

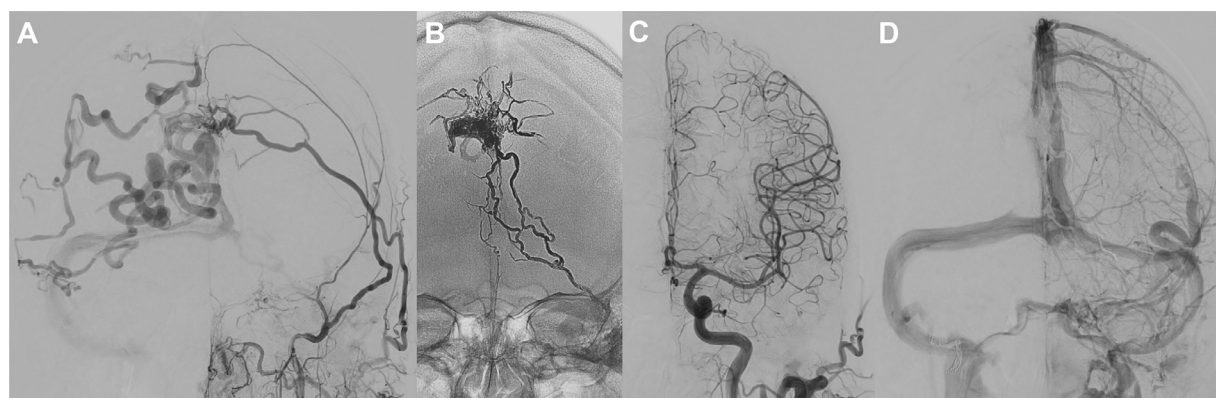


Fig. 4 – Angiography and endovascular treatment. (A) Arterial phase of initial angiography showing parasagittal dural arteriovenous fistula with severe corticovenous reflux. (B) Radiographic image showing the material injected by trans-arterial Onyx embolization. (C, D) Arterial and venous phase of final angiography showing elimination of arteriovenous shunt.

to a rehabilitation hospital 2 weeks after the intervention for residual left hemianopsia and mild higher brain dysfunction.

Discussion

We reported a case of unilateral subcortical calcification concomitant with cerebral edema, which subsequently developed into cerebral hemorrhage and was treated for an aggressive DAVF. The subcortical calcification concomitant with cerebral edema in patients with DAVF may suggest severe CVR, which is indicative of aggressive DAVF.

Unilateral subcortical calcifications with edema may indicate DAVF with severe CVR. These findings are typically observed in neoplasms prone to calcification [1,2]. Oligodendrogliomas, the initially suspected diagnosis in our case, are associated with calcification in up to 90% of cases, showing mixed findings of high- and low-density areas on CT [3,10]. Conversely, subcortical calcifications associated with

DAVF are not often accompanied by edema, or if present, are with only mild edema [4–6]. However, DAVF with severe CVR, although a small proportion of all DAVFs, may be associated with edema due to medullary venous congestion [11,12]. Therefore, the presence of both edema and calcification in a DAVF, while rare, may be an advanced manifestation of DAVF.

Calcifications in DAVF are thought to be caused by the chronic impairment of venous return [4,13,14]. In addition, edema can occur with extremely severe CVR, indicating a change in the venous drainage pattern due to a mechanism such as thrombosis of the drainage vein [11,15,16]. Several studies have shown that CVR is the most relevant factor for hemorrhagic events in DAVF [7,17,18]. Therefore, a DAVF with calcification and edema may be a dangerous condition, with a high likelihood of developing hemorrhage early after diagnosis, as seen in our case. This possibility that the presence of calcification alone and the presence of both calcification and edema may indicate a different disease stage warrants further investigation in a large number of cases.

Our case highlights the differential diagnosis of unilateral subcortical calcification with edema as a presenting feature of an aggressive DAVF. Thus, early evaluation of the cranial vessels is recommended for the diagnosis of DAVF.

Patient consent

Informed consent has been obtained from the patient's family member for publication of the case report and accompanying images.

REFERENCES

- [1] Grech R, Grech S, Mizzi A. Intracranial calcifications. A pictorial review. *Neuroradiol J* 2012;25:427–51. doi:[10.1177/197140091202500406](https://doi.org/10.1177/197140091202500406).
- [2] Saade C, Najem E, Asmar K, Salman R, El Achkar B, Naffaa L. Intracranial calcifications on CT: an updated review. *J Radiol Case Rep* 2019;13:1–18. doi:[10.3941/jrcr.v13i8.3633](https://doi.org/10.3941/jrcr.v13i8.3633).
- [3] Smits M. Imaging of oligodendroglioma. *Br J Radiol* 2016;89:20150857. doi:[10.1259/bjr.20150857](https://doi.org/10.1259/bjr.20150857).
- [4] Yang M-S, Chen CC-C, Cheng Y-Y, Yeh D-M, Lee S-K, Tyan Y-S. Unilateral subcortical calcification: a manifestation of dural arteriovenous fistula. *AJNR Am J Neuroradiol* 2005;26:1149–51.
- [5] Metoki T, Mugikura S, Higano S, Ezura M, Matsumoto Y, Hirayama K, et al. Subcortical calcification on CT in dural arteriovenous fistula with cortical venous reflux. *AJNR Am J Neuroradiol* 2006;27:1076–8.
- [6] Pascoe HM, Lui EH, Mitchell P, Gaillard F. Progressive subcortical calcifications secondary to venous hypertension in an intracranial dural arteriovenous fistula. *J Clin Neurosci* 2017;39:98–101. doi:[10.1016/j.jocn.2017.01.019](https://doi.org/10.1016/j.jocn.2017.01.019).
- [7] van Dijk JMC, terBrugge KG, Willinsky RA, Wallace MC. Clinical course of cranial dural arteriovenous fistulas with long-term persistent cortical venous reflux. *Stroke* 2002;33:1233–6. doi:[10.1161/01.str.0000014772.02908.44](https://doi.org/10.1161/01.str.0000014772.02908.44).
- [8] Gandhi D, Chen J, Pearl M, Huang J, Gemmete JJ, Kathuria S. Intracranial dural arteriovenous fistulas: Classification, imaging findings, and treatment. *AJNR Am J Neuroradiol* 2012;33:1007–13. doi:[10.3174/ajnr.A2798](https://doi.org/10.3174/ajnr.A2798).
- [9] Miller TR, Gandhi D. Intracranial dural arteriovenous fistulae: clinical presentation and management strategies. *Stroke* 2015;46:2017–25. doi:[10.1161/STROKEAHA.115.008228](https://doi.org/10.1161/STROKEAHA.115.008228).
- [10] Koeller KK, Rushing EJ. From the archives of the AFIP: oligodendroglioma and its variants: radiologic-pathologic correlation. *Radiographics* 2005;25:1669–88. doi:[10.1148/rq.256055137](https://doi.org/10.1148/rq.256055137).
- [11] Yu W, Rives J, Welch B, White J, Stehel E, Samson D. Hypoplasia or occlusion of the ipsilateral cranial venous drainage is associated with early fatal edema of middle cerebral artery infarction. *Stroke* 2009;40:3736–9. doi:[10.1161/STROKEAHA.109.563080](https://doi.org/10.1161/STROKEAHA.109.563080).
- [12] Lin Y-H, Lee C-W, Wang Y-F, Lu C-J, Chen Y-F, Liu H-M. Engorged medullary vein on CT angiography in patients with dural arteriovenous fistula: prevalence, types, and comparison between regional and extensive types. *J Neurointerv Surg* 2018;10:1114–19. doi:[10.1136/neurintsurg-2017-013660](https://doi.org/10.1136/neurintsurg-2017-013660).
- [13] Wang PY, Liu LH, Shen WC. Dural arteriovenous malformation of the transverse sinus with sinus occlusion: report of a case. *J Formos Med Assoc* 1992;91:102–5.
- [14] Lai PH, Chang MH, Liang HL, Pan HB, Yang CF. Unusual signs for dural arteriovenous fistulas with diffuse basal ganglia and cerebral calcification. *Zhonghua Yi Xue Za Zhi* 2000;63:329–33.
- [15] Dong Y, Cao W, Huang L, Li L, Zhang Y, Dong Q, et al. Ipsilateral intracranial edema associated with drainage patterns of dural arteriovenous fistula. *J Stroke Cerebrovasc Dis* 2014;23:1094–8. doi:[10.1016/j.jstrokecerebrovasdis.2013.09.016](https://doi.org/10.1016/j.jstrokecerebrovasdis.2013.09.016).
- [16] Copelan AZ, Krishnan A, Marin H, Silbergleit R. Dural Arteriovenous fistulas: a characteristic pattern of edema and enhancement of the medulla on MRI. *AJNR Am J Neuroradiol* 2018;39:238–44. doi:[10.3174/ajnr.A5460](https://doi.org/10.3174/ajnr.A5460).
- [17] Söderman M, Pavic L, Edner G, Holmin S, Andersson T. Natural history of dural arteriovenous shunts. *Stroke* 2008;39:1735–9. doi:[10.1161/STROKEAHA.107.506485](https://doi.org/10.1161/STROKEAHA.107.506485).
- [18] Strom RG, Botros JA, Refai D, Moran CJ, Cross DT 3rd, Chicoine MR, et al. Cranial dural arteriovenous fistulae: asymptomatic cortical venous drainage portends less aggressive clinical course. *Neurosurgery* 2009;64:241–7 discussion 247–8. doi:[10.1227/01.NEU.0000338066.30665.B2](https://doi.org/10.1227/01.NEU.0000338066.30665.B2).