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Original Article

Detection Rate and Radiological Features of Asymptomatic Intracranial Dural Arteriovenous Fistula: Analysis of Magnetic Resonance Imaging Data of 11745 Individuals in the Japanese Brain Check-Up System

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Objective: Asymptomatic intracranial dural arteriovenous fistula (DAVF) is a rare disease that is often undiagnosed before symptom onset. The present study aimed to examine the detection rate and radiological features of asymptomatic intracranial DAVF using brain MRI data obtained from the Japanese brain check-up system.
Methods: We retrospectively identified 11745 individuals who underwent brain MRI between January 2010 and December 2014. After a routine brain MRI screening, a definite diagnosis was made based on DSA. Data regarding sex, age, disease location, classification type, and treatment method were extracted from the system database and patients' medical records.
Results: Six individuals (0.05%; mean age, 61.0 ± 9.7 years) were diagnosed with definite intracranial DAVF. The intracranial DAVFs were located in the transverse sinus, confluence, and tentorial sinus in 2, 1, and 3 case(s), respectively. Cortical venous reflux was confirmed in four cases (66.7%), and none of the cases had intracranial hemorrhage or venous congestion. All cases had infratentorial lesions and two-thirds were Borden type II/III.
Conclusion: The detection rate of asymptomatic intracranial DAVF was 0.05% based on the analysis of MRI data from the brain check-up system. Low-flow shunt and tiny cortical venous reflux were likely missed on MRI.

Keywords ▶ detection rate, radiological features, asymptomatic, dural arteriovenous fistula, brain check-up system

Introduction

Emerging studies have yielded new findings regarding the pathophysiology and natural history of intracranial dural arteriovenous fistula (DAVF). Consequently, the diagnosis of intracranial DAVF has become more frequent, expedited owing to recent advances in diagnostic technology.[1,2] Furthermore, recent advances in neuroradiology, especially DSA and cone-beam CT, have improved our understanding of this disease. In addition, improvements in endovascular devices have allowed endovascular therapy to become the primary treatment method for intracranial DAVF, resulting in more cases being successfully cured. However, the underlying pathophysiology and clinical features of intracranial DAVF remain unclear.

Intracranial DAVF is a dynamic disease with a chronological progression.[3] The natural history of intracranial DAVF has been considerably elucidated by retrospective cohort studies.[3–6] Intracranial DAVF is rare, with several reports showing that its incidence is 0.16–0.51 per 100000 adults per year.[7–9] These results are calculated based on autopsy cases and confirmed diagnosis data. Occasionally, incidental intracranial DAVF can emerge, while cases can

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also undergo spontaneous regression. Therefore, it is possible that the prevalence of intracranial DAVF is higher than expected. Establishing the true prevalence of intracranial DAVF will clarify its natural history and permit the formulation of more appropriate criteria for treatment indications and methods. However, since intracranial DAVF is rare, it is difficult to investigate its frequency and prevalence in the general population.

In Japan, any person can pay to undergo MRI and MRA to check the health of their brain through the brain check-up system.[10] Therefore, this system can be used to evaluate the frequency of rare diseases.[11] This study aimed to examine the detection rate of asymptomatic intracranial DAVF based on data from the Japanese brain check-up system. To the best of our knowledge, no previous study has reported the detection rate and radiological features of asymptomatic intracranial DAVF based on MRI. We believe that this study offers valuable information that may help to elucidate the pathophysiology of intracranial DAVF.

Materials and Methods

Study population

In this single-center, cross-sectional study, we performed a retrospective analysis of MRI data from the brain check-up system, which is a check-up system for neurological diseases in Japan. This system includes MRI and MRA data of the brain, carotid ultrasound, and an assessment of the individual's mental status, in addition to regular physical and laboratory findings. Ordinary people can utilize the brain check-up system to check their brain health at their own or their employer's expense. The cost ranges from 300 to 1000 US dollars.[10] A total of 11745 individuals who underwent brain MRI between January 2010 and December 2014 were eligible for the study. In cases where multiple MRI scans were performed on the same subject within the study period, we evaluated only the first MRI scan. The clinical presentation was considered asymptomatic if the person had no subjective complaints, headache, pulsatile tinnitus, or neurological deficits. As most subjects were middle-aged to older individuals, this study was unable to investigate the detection rate of asymptomatic intracranial DAVF in younger people.

MRI and diagnosis

The brain check-up involved performing MRI and MRA on all subjects using a 3.0 Tesla superconducting system (Achieva 3.0T X-series; Philips Healthcare, Best, The Netherlands). Except for time-of-flight MRA (TOF-MRA), which scanned the area from the cranio-cervical junction to the body of the lateral ventricle, the rest of the tests scanned the entire head. At the time of examination, experienced board-certified neurosurgeons or neurologists checked the MRI scans. Subsequently, different board-certified, experienced neurosurgeons rechecked the MRI scans based on the diagnosis by the first evaluator. If unusual findings were observed on the scans, the individual was referred to a specialized hospital for additional examinations. A probable diagnosis of intracranial DAVF was made based on the following criteria: high-intensity areas in the venous sinus or intracranial veins on TOF-MRA and high-intensity signals originating from feeders at the sinus wall in the TOF-MRA source images.[2] Finally, a definite diagnosis was made by an experienced neurosurgeon at a specialized hospital using DSA. We also retrieved information from the brain check-up system database and medical records on the individual's age, sex, location of the DAVF, Borden classification,[12] Cognard classification,[13] presence/absence of cortical venous reflux, symptoms, and treatment.

Given the anonymous nature of the data, the requirement for informed consent was waived. This study was approved by the institutional review board of our hospital.

Statistical analysis

Continuous data are expressed as the mean ± standard deviation while discrete data are presented as the number and percentage. We calculated the detection rate of intracranial DAVF using the cohort of all individuals with records in the brain check-up system. We calculated the 95% confidence intervals for detection rate estimates using a Poisson distribution. Analyses were performed using SPSS software (version 22; IBM, Armonk, NY, USA).

Results

A total of 11745 individuals (6980 men and 4765 women), with a mean age of 56.7 ± 10.0 years (range, 41–80 years), were eligible for the study. Based on data from the brain check-up system, 14 (0.12%) individuals were diagnosed with probable arteriovenous shunt disease, 7 (0.06%) of whom were diagnosed with probable intracranial DAVF, and they subsequently underwent DSA. The additional examination resulted in 6 (0.05%) individuals being diagnosed with definite intracranial DAVF (**Table 1**) (Case 2, **Fig. 1**) (Case 4, **Fig. 2**). The mean age of these individuals was 61.0 ± 9.7 years, and 4 (66.7%) of them were men. In

Table 1 Summary of the intracranial dural arteriovenous fistula cases

Case	Subject age, sex	Site	Borden	Cognard	CVR	Clinical course
1	63, Male	Transverse sigmoid	III	III	+	TAE
2	72, Female	Transverse sigmoid	I	IIa	−	Spontaneous resolution
3	58, Male	Tentorial	II	IIa + b	+	TAE + TVE
4	46, Male	Tentorial	III	III	+	TAE
5	55, Male	Tentorial	II	IIa + b	+	TVE
6	74, Female	Confluence	I	I	−	Follow up

CVR: cortical venous reflux; TAE: transarterial embolization; TVE: transvenous embolization

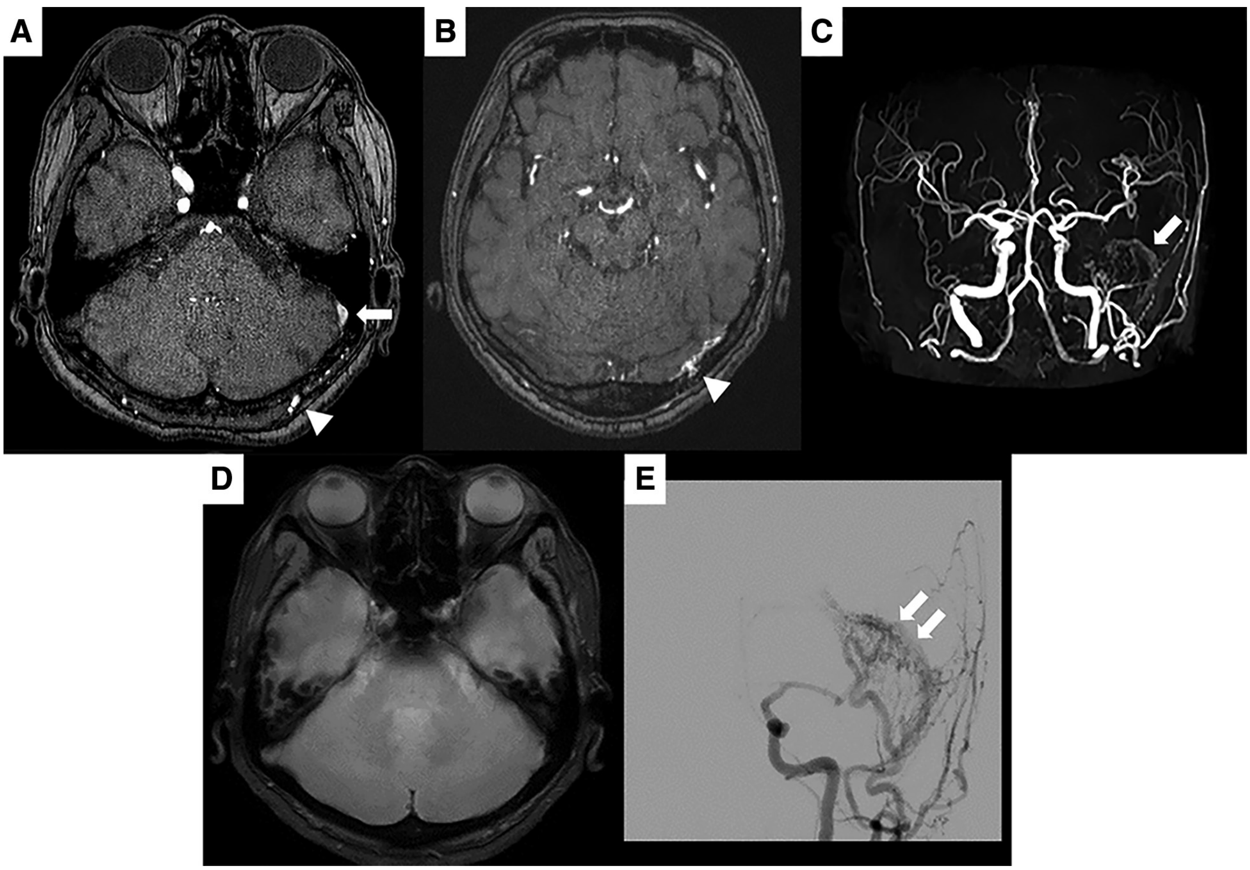

Fig. 1 Images from the patient in Case 2. The patient is a 72-year-old woman with a transverse-sigmoid DAVF (Cognard IIa). (**A–C**) TOF-MRA and MRA images from the medical check-up system reveal high-intensity areas in the transverse sigmoid (single arrows) and high-intensity signals originating from dilated feeders (arrowheads). (**D**) MR T2*-weighted image shows no hemorrhagic findings. (**E**) Anteroposterior view of the left common carotid arteriogram shows that the occipital artery mainly forms the DAVF in the TSS without cortical venous reflux (double arrows). DAVF: dural arteriovenous fistula; TOF: time-of-flight; TSS: transverse sigmoid sinus

all cases, lesions occurred in the posterior cranial fossa. Additionally, lesions occurred in the transverse sinus, confluence, and tentorial sinus in 2, 1, and 3 case(s), respectively. Cortical venous reflux was confirmed in four cases (66.7%). Based on the Borden classification, two cases were classified as type I lesions, two as type II lesions, and two as type III lesions. Based on the Cognard classification, one case was classified as type I, one as type II, two as type IIa + b, and two as type III. No parenchymal changes were observed in any of the cases. All cases were asymptomatic, and none of the assessed individuals had undergone craniotomy.

Treatment was performed on four patients with cortical venous reflux; among them, two patients did not require further treatment. The intracranial DAVF in one patient (Case 2) resolved spontaneously. The other patient (Case 6) did not have cortical venous reflux and was asymptomatic; therefore, imaging was subsequently performed.

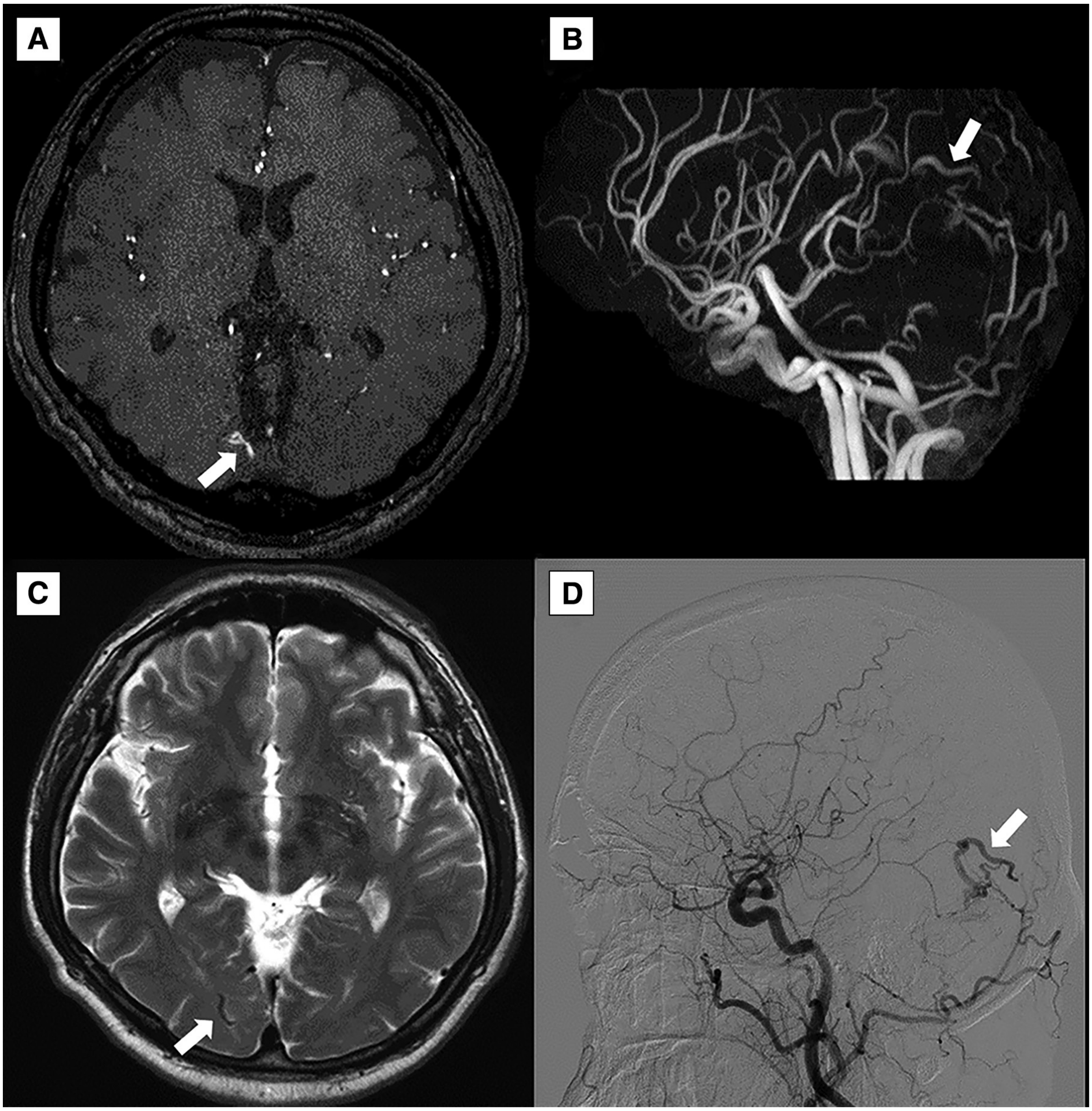

Fig. 2 Images from the patient in Case 4. The patient is a 46-year-old man with a tentorial DAVF (Cognard III). (**A**) TOF-MRA image reveals dilated cortical veins (arrow). (**B**) MRA image reveals high-intensity areas in the cortical vein (arrow). (C) MR T2-weighted image shows a dilated cortical vein (arrow). (**D**) Lateral view of the common carotid arteriogram shows that the artery of the falx cerebelli primarily forms the tentorial DAVF (arrow). DAVF: dural arteriovenous fistula; TOF: time-of-flight

Discussion

This study used MRI data from the Japanese brain check-up system to investigate the detection rate and radiological features of asymptomatic/incidental intracranial DAVF. Among the 11745 individuals who underwent their brain check-up, 6 (0.05%) individuals were diagnosed with intracranial DAVF. In all cases, the DAVF lesions were located in the posterior cranial fossa. Among the six individuals with intracranial DAVF, four (66.7%) had cortical venous reflux, and no parenchymal changes were observed in any of them. Our detection rate of asymptomatic intracranial DAVF based on brain check-up MRI was 10.2 per 100000 individuals per year. The results of this study differed greatly from those reported previously, as follows: (1) detection rate was much higher, (2) infratentorial lesions were more common, and (3) the prevalence of cortical venous reflux was higher.

Previous retrospective studies have reported the incidence of intracranial DAVF as 0.15–0.51/100000 individuals per year.[7–9] There are several potential causes for the increase in frequency revealed by this study. First, the previous reports utilized data from confirmed diagnosis and autopsy cases to establish the background population; thus, potential and indefinite cases were not included. In this study, since the subjects were ordinary people who had undergone examinations as part of the Japanese brain check-up system, potential and asymptomatic lesions were detected. Additionally, neuroradiological technology has been improving, possibly resulting in a greater detection rate. MRI/MRA has a good diagnostic accuracy for the detection of cortical venous reflux in patients with intracranial DAVFs.[14)] Regardless, it is difficult to determine the presence of an arteriovenous shunt even with independent confirmation by two neurologically trained clinicians, such as in this study. The true prevalence of this condition is considered higher than what the present data suggest because MRI/MRA cannot detect all cases. It might be especially difficult to detect DAVFs in cases with small shunt volume and inconspicuous cortical venous reflux. Previous reports showed the efficacy of time-resolved contrast-enhanced MRA and CTA techniques, which have high sensitivity and specificity for the detection of DAVFs.[14,15)] Furthermore, arterial spin-labeling is a new method of noninvasive MRI that can detect intracranial DAVF.[16)] However, these techniques have disadvantages, including the side effects of contrast agents, radiation exposure, additional time, and low cost-effectiveness. In addition, DAVFs, which are hard to detect by MRI/MRA, have small shunt volume and cortical venous reflux; thus, their clinical significance might be considerably low.

Herein, four (66.7%) of the six individuals with asymptomatic intracranial DAVF had cortical venous reflux. A retrospective study of 251 patients with symptomatic DAVF in Finland showed that cortical venous reflux was present in 88 fistulas (34%).[8)] Although there were fewer individuals with intracranial DAVF in our study, the occurrence of cortical venous reflux was more frequent than that reported previously. Cortical venous reflux influences the classification and treatment strategy for DAVF because aggressive types of DAVF—those involving hemorrhage and neurological deficits—are caused by cortical venous reflux. In contrast, some studies have reported that cases of intracranial DAVF with asymptomatic cortical venous reflux have a less aggressive clinical course than those with symptomatic cortical venous reflux.[3–5)] An observational follow-up study of 112 patients reported that only 4% of the patients showed conversion to an aggressive lesion from Boden type I to type II/III.[3)] Therefore, it is estimated that cortical venous reflux might be present from the early stage rather than with stage progression, and cortical venous reflux might be present in asymptomatic cases.

Interestingly, all cases in this study had infratentorial lesions. However, a previous study reported that cavernous sinus (CS) lesions (43.6%) and transverse sigmoid sinus (TSS) lesions (33.4%) were the most common in the Japanese population.[17)] Previous studies from Western countries have reported that DAVFs are most frequently located in the TSS, followed by the CS.[8,13)] CS DAVFs were not identified in this study. Patients with CS DAVFs easily become symptomatic because the CS is located close to the eyes and cranial nerves, including the oculomotor, trochlear, and abducens nerves. Unlike infratentorial DAVFs, CS DAVFs can become symptomatic with just sinus and cortical venous reflux. Infratentorial DAVFs reportedly become symptomatic after intracranial hemorrhage, remarkable cortical venous reflux, and cerebral edema. These results suggest that there may be more asymptomatic infratentorial DAVFs.

There are several limitations to our study that should be acknowledged. First, cases that did not show abnormal MRA findings might have led to missed diagnoses. Occasionally, the diagnosis of TSS DAVF with MRA alone was difficult, and superior sagittal sinus DAVF may have been overlooked because it was located outside of the MRA scan area. Second, there may have been selection bias since the assessments were performed at the examinee's own expense (about 300–1000 US dollars). Moreover, children, young adults, and older adults did not undergo medical check-ups with brain MRI (mean age of the study participants, 56.7 ± 10.0 years; range, 41–80 years); people who undergo such brain check-ups tend to have financial reserves for healthcare; therefore, this study did not reflect the actual population ratio for the disease occurrence.

Conclusion

In conclusion, the detection rate of asymptomatic/incidental intracranial DAVF in our study utilizing the brain check-up system was 0.05%, with all cases presenting infratentorial lesions of which two-thirds were Borden type II/III. However, the detection of low-flow shunt and tiny cortical venous reflux was likely missed on MRI; thus, their detection rate remains elusive. These results might help clinicians to accurately diagnose intracranial DAVFs. Further study is warranted to establish the pathophysiology of intracranial DAVF.

Disclosure Statement

The authors declare that there is no conflict of interest.

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