

CASE REPORT

Case report: Migraine that persisted for over 20 years disappears after treatment for pulmonary arteriovenous fistula

Kazuyuki Kakeshita¹ | Taro Yoneda¹  | Hayato Koba¹ | Kota Tanimura¹ | Tsukasa Ueda¹ | Tomoya Kaneda¹ | Johsuke Hara² | Kazuo Kasahara²

¹Department of Internal Medicine, Komatsu Municipal Hospital, Ishikawa, Japan

²Department of Respiratory Medicine, Kanazawa University Hospital, Ishikawa, Japan

Correspondence

Taro Yoneda, Department of Internal Medicine, Komatsu Municipal Hospital, Ishikawa, Ho-60 Mukaimotoorimachi, Komatsu, Ishikawa 923-8560, Japan.
Email: taroyoneda@yahoo.co.jp

Abstract

We presented a rare case of pulmonary arteriovenous fistula in a patient who suffered from migraine with optic aura for longer than 20 years. This case suggests that the migraine could be expected to disappear after treatment for pulmonary arteriovenous fistula.

KEY WORDS

cerebral infarction, migraine, pulmonary arteriovenous fistula, shunt, transesophageal echocardiography

1 | INTRODUCTION

Pulmonary arteriovenous fistula (PAVF) is an important disease state that is considered a risk factor of cerebral infarction.¹ A high incidence (38.5%) of migraine with visual aura concomitant with PAVF has been reported.² However, a case of migraine, which had persisted for more than 20 years, disappearing directly following PAVF treatment has not yet been reported. We experienced a case where this phenomenon occurred.

Here, we describe the clinical course of a patient with cerebral infarction resulting from PAVF; our examination suggested that this patient's migraine of unknown cause disappeared upon treating the PAVF.

2 | CASE REPORT

A 41-year-old woman presented to our hospital in 2017. She has a history of migraine with visual aura once or twice every month for more than 20 years and recurrent cerebral infarction between the ages of 32-37. At that time, it was thought

that the cerebral infarction derived from migraine. This time, she lost right visual field after urinating upon waking and had loss of muscle strength in her right upper and lower extremities. Her visual field loss continued for 5 minutes accompanied with left occipital pain. Because the loss of muscle strength of her right upper and lower extremities persisted, she presented to the emergency department of our hospital. She had a clear sensorium and normal respiratory sound, with percutaneous oxygen saturation at 97%. Diffusion-weighted brain magnetic resonance imaging (MRI) showed high signal in the left backward lobus temporalis, which indicated fresh cerebral infarction (Figure 1). In the past, she had magnetic resonance angiography (MRA) which had never shown vascular abnormality.

Repeated MRI also showed no abnormality. Upon admission, her right upper and lower extremities had loss of muscle strength with level 4 rating on manual muscle test, but returned to normal on the same day, and she had no sensory impairment. Additional blood examination showed no evidence of angitis and antiphospholipid antibody syndrome (APS). Examination of cerebrospinal fluid was performed, and the results were negative for the possibility

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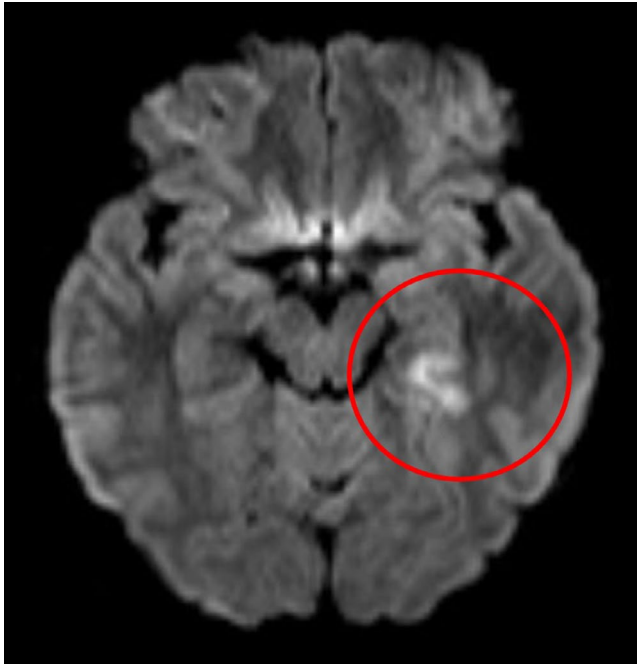


FIGURE 1 Diffusion-weighted imaging of brain magnetic resonance imaging showed high signal in left backward lobe temporalis, which indicated fresh cerebral infarction (inside red circle)

of demyelinating disorder, such as meningitis (Table 1). Nevertheless, chest X-ray showed a 1-cm node in the left lower lung.

To search for cause of the cerebral infarction, transesophageal echocardiography (TEE) was performed. Bubble contrast method showed no interatrial access. Nevertheless, there was a bubble lag time of about 1 or 2 breaths between the right and left atria (Figure 2), which indicated an existence of shunt in the atrium. Because PAVF was strongly suspected, an enhanced chest computed tomography (CT) was performed and showed PAVF which was 1.5 cm in the peripheral left superior lingular segment (Figure 3). Visible vessel of gastric mucosa was not confirmed with upper gastrointestinal endoscopy. The patient was not prone to nosebleed and there was no family history of hereditary telangiectasia, which can be diagnosed based on the presence of 4 criteria, known as the “Curaçao criteria”.³ As previously mentioned, we thought that this cerebral infarction had been derived from idiopathic PAVF as paradoxical embolism. Thus, left upper lobe partial resection was performed to treat PAVF. Macroscopically, we found blood blister-like dilated vessels on the visceral pleura surface outside the left superior lingular segment, and a 17-mm PAVF formed by aggregation of the dilated vessels was confirmed in the peripheral superior lingular segment (Figure 4). On postoperative day 33, transesophageal echocardiography was performed once again, and the shunt disappeared. After discharge from the hospital, chest CT did not show new PAVF. At the time this happened, she had been visiting a doctor regularly for 2 years. Her migraine, which

would develop 1 or 2 times every month before the treatment of PAVF, completely stopped. It was thought that her migraine with optic aura which persisted for over 20 years derived from PAVF.

3 | DISCUSSION

Pulmonary arteriovenous fistula is congenital in more than 90% of cases. Approximately 50% of congenital PAVF in the United States and 15% of congenital PAVF in Japan were complicated with hereditary telangiectasia,⁴ and 15% of hereditary telangiectasia was complicated with pulmonary AVF. In other words, when we encounter the diagnosis of PAVF, we should also take into consideration hereditary telangiectasia. It is reported that 9%–41% of central neurologic disease, such as cerebral stroke and brain abscess, was due to PAVF. In Japan, 0.5% of patients with recurrent cerebral infarction (4 of 754 cases) were reported to be caused by PAVF.¹ Six years after the diagnosis of PAVF, the fatality rate was reported to be 11%. Therefore, aggressive management should be taken into consideration. Cases that present with hypoxemia, cardiac failure, central nervous system manifestation, and inflow artery diameter over 2–3 mm indicate treatment for PAVF,⁵ which includes surgical resection, such as segmental resection, excision of fistula, or transcatheter embolization. Surgical resection was performed for giant fistula or artery with large diameter of inflow in which blood current would stick. Currently, transcatheter embolization has become mainstream because of low invasiveness, low complication, and the ability to be performed with another embolization.⁶ It is reported that transcatheter embolization could be considered in the prevention of relapse of cerebral infarction caused by PAVF.¹ Transcatheter embolization less invasively achieves 98% of treatment of PAVF and improves hypoxemia and hypoxemia. Nevertheless, it was reported that 1%–10% of cases with transcatheter embolization were recanalized in the long term.⁵ On the other hand, surgical resection was reported as definitive therapy whose outcome is unassociated with size of pathologic lesion.

Minimally invasive thoracoscopic surgery has evolved in recent years to provide reliable and definitive repair in patients who can be given general anesthetic.⁷

In our case, surgical resection was chosen by the patient, because she was young and hoped for definitive repair. PAVF is an important disease state considered a risk factor of cerebral infarction as mentioned previously. It is noteworthy in this case that migraine was reported to coexist with PAVF and patent foramen ovale. Migraine with visual aura is recognized to coexist with PAVF in 38.5% of cases.² In addition, it has been reported that migraine

TABLE 1 Laboratory findings upon admission

Blood				Blood gas analysis (room air)	
WBC	7900 / μ L	PIC	0.5 μ g/mL	pH	7.402
Neut	50.4%	TAT	2.6 ng/mL	pO ₂	98.3 Torr
Eos	3.2%	Plasminogen	114%	pCO ₂	37.5 Torr
Lym	36.9%	Protein C	128%	HCO ₃ ⁻	22.9 mEq/L
RBC	432 $\times 10^4$ / μ L	Protein S	88%	SaO ₂	97.5%
Hb	13.2 g/dL	Lupus anticoagulant	Negative	A-aDO ₂	9.8 Torr
MCV	91.1%	Anticardiolipin antibody	8 U/mL	Cerebrospinal fluid examination	
Plt	29.5 $\times 10^4$ / μ L	IgG	1057 mg/dL	Initial pressure	11 cmH ₂ O
Na	137 mEq/L	IgA	171 mg/dL	Appearance	
CL	102 mEq/L	IgM	121 mg/dL	Colorless and transparent	
K	3.8 mEq/L	IgE	18.8 U/mL	Specific gravity	1.005
AST	26 U/L	C3	110 mg/dL	pH	7.6
ALT	33 U/L	C4	21.7 mg/dL	Sugar	66.0 mg/dL
TP	6.6 g/dL	RF	15 U/mL	Cell number	Mononuclear 1 / μ L
Alb	4.0 g/dL	Antinuclear antibody	<40		Polynuclear 1 / μ L
LDH	141 U/L	Anti SS-A antibody	<1.0 U/mL	Protein	18 mg/d
BUN	15.4 U/L	Anti SS-B antibody	1.1 U/mL	CL	124 mEq/L
Cr	0.41 mg/dL	PR3-ANCA	<1.0 U/mL	Myelin basic protein	40.0 pg/mL
CRP	0.08 mg/dL	MPO-ANCA	<1.0 U/mL	Oligoclonal band	Negative
BS	107 mg/dL	Cryoglobulin	Negative	IgG index	0.51
PT	99.00%				
PT-INR	1.01				
APTT	31.40 s				
Fib	212 mg/dL				
D-dimer	1.0 μ g/mL				

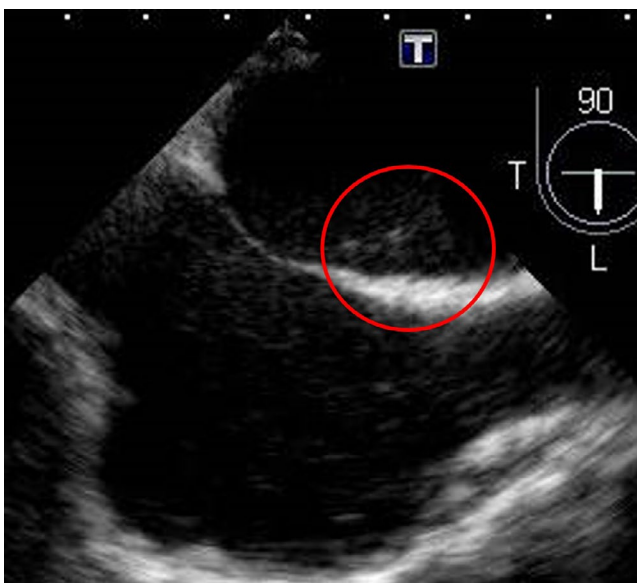
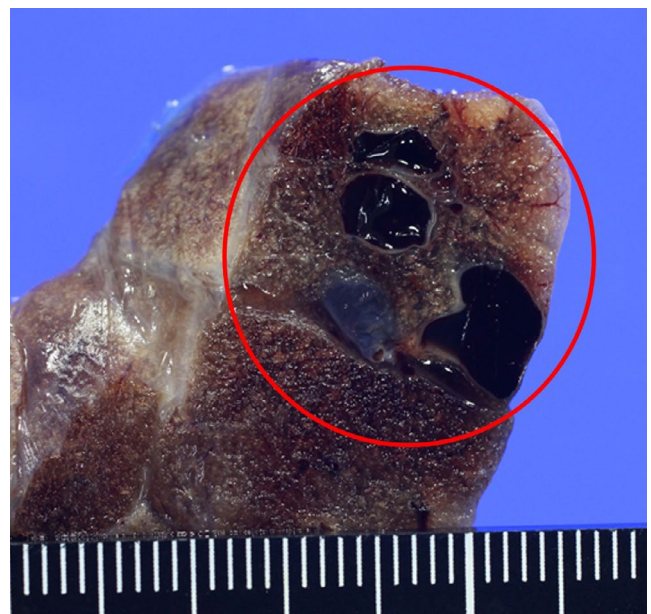
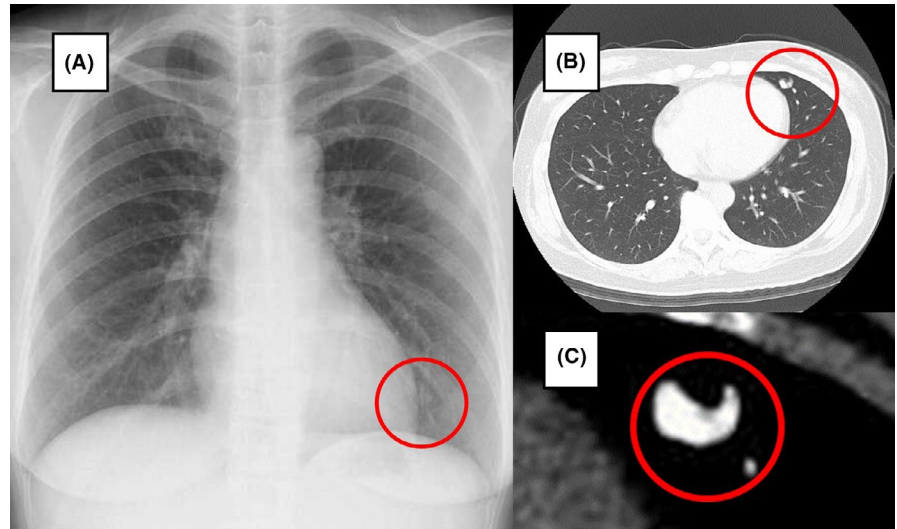
**FIGURE 2** On transesophageal echocardiography, bubble contrast method showed time lag of the bubble about one or two breaths between the right and left atria**FIGURE 4** Resected specimen of pulmonary AVF which was 17 mm long formed by aggregation of the dilated vessels

FIGURE 3 Chest X-ray (A) and the enhanced computed tomography (B,C) showed pulmonary AVF which was 1.5 cm in the peripheral left superior lingular segment which was contrasted (C, inside red circle)



stopped after treatment of PAVF and patent foramen ovale.^{8,9} Furthermore, the rate of migraine improvement was over 90% after 3 months of patent foramen ovale closure in patients with large-diameter patent foramen ovale complicated by migraine.¹⁰

Nevertheless, prospective tests showed no remarkable differences in improvement of migraine.¹¹⁻¹³ It was reported that migraine would stop after transcatheter closure of atrial septal defect.¹⁴ The mechanism of migraine improvement due to treatment of PAVF was not accurately known. Migraine with shunt is common in PAVF, patent foramen ovale, and atrial septal defect. It has been reported that migraine may occur from the inflow of serotonin and endothelin from venous system into arterial system via shunt.¹⁵ This same mechanism could be at work in pulmonary PAVF.

Transesophageal echocardiography is useful for detecting left atrial appendage thrombus, and left and right shunt when compared with transthoracic echocardiography.

Transesophageal echocardiography has been a prominent in the search for the cause of cerebral infarction.¹⁶ Likewise, in our case, TEE was an important examination.

In conclusion, in our case, PAVF was found when we searched for the cause of recurrent cerebral infarction in a patient who suffered from migraine with visual aura for more than 20 years. With the treatment of PAVF to prevent recurrent cerebral infarction, the sustained migraine stopped and disappeared.

While the cause of migraine with visual aura remains to be identified, we should take into consideration PAVF as cause of the migraine. Transthoracic echocardiography is very useful to detect shunt disease; however, in this case we used TEE. Once the diagnosis of PAVF is made in a patient with migraine, the migraine could be expected to disappear after treatment of PAVF.

CONFLICT OF INTEREST

None declared.

AUTHOR CONTRIBUTIONS

KK, HK, KT, and TU: drafted the contribution. TY: drafted, reviewed, edited, and conceived the contribution. TK, JH, and KK: conceived the contribution.

ORCID

Taro Yoneda  <https://orcid.org/0000-0002-7990-4058>

REFERENCES

- Kimura K, Minematsu K, Nakajima M. Isolated pulmonary arteriovenous fistula without Rendu-Osler-Weber disease as a cause of cryptogenic stroke. *J Neurol Neurosurg Psychiatry*. 2004;75:311-313.
- Marziniak M, Jung A, Guralnik V, et al. An association of migraine with hereditary haemorrhagic telangiectasia independently of pulmonary right-to-left shunts. *Cephalalgia*. 2009;29:76-81.
- Shovlin CL, Guttmacher AE, Buscarini E, et al. Diagnostic criteria for hereditary hemorrhagic telangiectasia (Rendu-Osler-Weber syndrome). *Am J Med Genet*. 2000;91:66-67.
- Swanson KL, Prakash UB, Stanson AW. Pulmonary arteriovenous fistulas: Mayo Clinic experience, 1982-1997. *Mayo Clin Proc*. 1999;74(7):671-680.
- Pollak JS, Saluja S, Thabet A, et al. Clinical and anatomic outcomes after embolotherapy of pulmonary arteriovenous malformations. *J Vasc Interv Radiol*. 2006;17(1):35-44.
- White RI, Lynch-Nyhan A, Terry P, et al. Pulmonary arteriovenous malformations: techniques and long-term outcome of embolotherapy. *Radiology*. 1988;169(3):663-699.
- Watanabe N, Hirayama J, Munakata Y, et al. A case of pulmonary arteriovenous malformation in a patient with brain abscess successfully treated with video-assisted thoracoscopic resection. *Chest*. 1995;108:1724-1727.
- Post MC, Thijs V, Schonewille WJ, et al. Embolization of pulmonary arteriovenous malformations and decrease in prevalence of migraine. *Neurology*. 2006;66:202-205.

9. Morandi E, Anzola GP, Angeli S, et al. Transcatheter closure of patent foramen ovale: a new migraine treatment? *J Interv Cardiol.* 2003;16:39-42.
10. Takaya Y, Akagi T, Kijima Y, et al. Influence of transcatheter closure of atrial communication on migraine headache in patients with ischemic stroke. *Cardiovasc Interv Ther.* 2016;31(4):263-268.
11. Dowson A, Mullen MJ, Peatfield R, et al. Migraine Intervention With STARFlex Technology (MIST) trial: a prospective, multicenter, double-blind, sham-controlled trial to evaluate the effectiveness of patent foramen ovale closure with STARFlex septal repair implant to resolve refractory migraine headache. *Circulation.* 2008;117(11):1397-1404.
12. Mattle HP, Evers S, Hildick-Smith D, et al. Percutaneous closure of patent foramen ovale in migraine with aura, a randomized controlled trial. *Eur Heart J.* 2016;37:2029-2036.
13. Charles A, Silberstein S, Sorensen S, et al. Results of the PREMIUM trial: patent foramen ovale closure with the AMPLATZ™ PFO occluder for the prevention of migraine. *Headache.* 2015;55(S5):251.
14. Kato Y, Kobayashi T, Ishido H, et al. Migraine attacks after transcatheter closure of atrial septal defect has also been reported. *Cephalalgia.* 2013;33(15):1229-1237.
15. Borgdorf P, Tangelder GJ. Migraine: possible role of shear-induced platelet aggregation with serotonin release. *Headache.* 2012;52:1298-1318.
16. Hashimoto Y, Terashima S. Diagnosis of cardiogenic cerebral embolism. *Adv Neurol Sci.* 2001;45(3):437-449.

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