ScientificScholar®
Knowledge is power
Publisher of Scientific Journals

www.surgicalneurologyint.com

Surgical Neurology International

Editor-in-Chief: Nancy E. Epstein, MD, Clinical Professor of Neurological Surgery, School of Medicine, State U. of NY at Stony Brook.

SNI: Unique Case Observations

Editor
S. Ather Enam, MD, PhD
Aga Khan University, Karachi, Sindh, Pakistan

SNI
Open Access

Case Report

The first 3-D volumetric analysis of mesencephalothalamic giant perivascular spaces showing steady and slow growth over 17 years

Tomohisa Okada[1], Kaisei Makimoto[2], Kayoko Itoh[3], FM Moinuddin[4], Koji Yoshimoto[5], Kazunori Arita[1,5]

[1]Department of Neurosurgery, Izumi Regional Hospital, Kagoshima, Japan, [2]Department of Radiology, Izumi Regional Hospital, Kagoshima, Japan, [3]Department of Neurology, Uchiyama Hospital, Akune, Kagoshima, Japan, [4]Department of Neurologic Surgery, Mayo Clinic, Rochester, Minnesota, United States, [5]Department of Neurosurgery, Kagoshima University, Sakuragaoka, Kagoshima, Japan.

E-mail: Tomohisa Okada - okadatomohisa794@gmail.com; Kaisei Makimoto - spector09335rt@gmail.com; Kayoko Itoh - kayo110@citrus.ocn.ne.jp; FM Moinuddin - moin_mmc35@yahoo.com; Koji Yoshimoto - kyoshimo@m.kufm.kagoshima-u.ac.jp; *Kazunori Arita - karita@m2.kufm.kagoshima-u.ac.jp

***Corresponding author:**
Kazunori Arita,
Emeritus Professor, Department of Neurosurgery, Kagoshima University, Sakuragaoka, Kagoshima, Japan.

karita@m2.kufm.kagoshima-u.ac.jp

Received : 11 July 2020
Accepted : 01 September 2020
Published : 18 September 2020

DOI
10.25259/SNI_423_2020

Quick Response Code:

ABSTRACT

Background: Giant perivascular spaces (PVSs) are very rare condition in the brain and can be associated with neurological symptoms. It often enlarges and causes obstructive hydrocephalus which requires surgical intervention. However, the growth velocity has never been investigated.

Case Description: Here, we report a woman in her early eighties with giant PVSs eventually followed up 17 years. She presented with dizziness and mild headache for a week and her neurological examination showed no abnormality. Her brain magnetic resonance imaging (MRI) showed a multiple cystic lesion, 28 mm in maximum diameter as a whole, in the left mesencephalothalamic region. There were no solid part, rim enhancement, or peri-lesional intensity change suggesting edema or gliosis. Smaller PVSs were also seen in bilateral-hippocampi, basal ganglia, white matter, and left frontal operculum. Retrospectively, five MRI studies over 17 years were analyzed using a 3-D volumetric software and found a very slow growth of the lesion, from 6.54 ml to 9.83 ml indicating gain of 0.1752 ml (2.68%) per year.

Conclusion: This is the first report verifying a gradual enlargement of giant PVSs in a natural course. The prospective 3-D volumetric analysis on PVSs may elucidate the true nature of these lesions.

Keywords: Giant perivascular spaces, Growth, Midbrain, Thalamus

INTRODUCTION

Perivascular spaces (PVSs) or Virchow-Robin spaces are pial-lined structures surrounding the vessels filled with interstitial/extracellular fluid. They are believed to serve as a lymphatic drainage pathway in the brain and play a significant role in the immune system in the brain.[2,12,13] With advancing age, they are found more frequently by magnetic resonance imaging (MRI).[4,7]

PVSs are more commonly seen in three locations.[5,10] Type I PVSs, most common, are surrounding lenticulostriate arteries entering the basal ganglia through the anterior perforated substance. Type II is found in subcortical white matter along perforating medullary arteries which penetrate the high-convexity gray matter. Type III appears in the midbrain/cerebral

peduncle and surrounds the penetrating branches of the paramedial mesencephalothalamic arteries. However, PVSs can be seen in any part of brain, such as temporal lobe, insula, and hippocampus. These PVSs sometimes become dilated very large as a single cyst or cluster of cysts of various sizes. When the size of PVSs is larger than 1.5 cm, it is called giant or tumefactive PVSs and occasionally causing mass effect.[10,11] These giant PVSs are often misdiagnosed as cystic neoplasm, non-neoplastic neuroepithelial cyst, or parasitic cyst. Here, we present a rare case of giant PVSs in the mesencephalothalamic region with 17 years follow-up showing gradual enlargement of the cyst.

CASE REPORT

An woman in her early eighties was referred to the department of neurosurgery, Izumi Regional Hospital, Japan from a neurologist on June 2020. She was a known case of cystic brain lesion since 2003. She presented with mild headache and dizziness for a week. The patient's history included diabetes and hypertension over 20 years. No abnormality was found on neurological examination including eye movement or cerebellar function.

MRI on June 2020 showed a cluster of cystic lesion, 28 mm in diameter as whole, in the left mesencephalothalamic region [Figure 1a]. The lesion was extended from the pontomesencephalic junction to the dorsal surface of pulvinar of thalamus. The cysts' content was hypointense on T1-weighted and fluid-attenuated inversion recovery (FLAIR) MRI and hyperintense on T2-weighted MRI [Figure 1b-g]. There was no hyperintensity on FLAIR surrounding the lesion suggesting edema or gliosis. The similarity of MR intensity of cyst content to cerebral fluid on all MR sequences, lack of perilesional edema, and absence of gadolinium enhancement strongly suggested this lesion to be asymptomatic giant PVSs. Smaller PVSs were seen in other brain regions including bilateral-hippocampi, basal ganglia, white matter under convexity gray matter, and left frontal operculum [Figure 2a-d]. T2-weighted and FLAIR MRI also showed age-matched brain atrophy and hyperintensity in periventricular and subcortical white matter suggesting leukoaraiosis. Brain and neck MR angiography showed no particular abnormality (figure not shown). Retrospective review of the medical record and image archives found four previous MR studies on September 2003, May 2014, February 2016, and July 2017 [Figure 3a-e] in our and other hospitals due to chronic headache and uneasiness caused by suggestion of brain tumor by a previous attending. 3D volumetric analysis of the giant PVSs using Synapse Vincent software (Fuji film, Tokyo) showed a very slow enlargement, 6.54 ml in September 2003–9.83 ml in June 2020. It has grown with slope of linear function: y (ml) = 0.1752x+6.0012 (x: year-2000), $R^2 = 0.9072$ [Figure 3f].

The patient is now scheduled to have another MRI study 2 years later for early detection of symptomatic enlargement including obstructive hydrocephalus resulting from aqueductal stenosis.

DISCUSSION

Giant PVSs or tumefactive PVSs are very rare lesion in the brain commonly found in mesencephalothalamic region

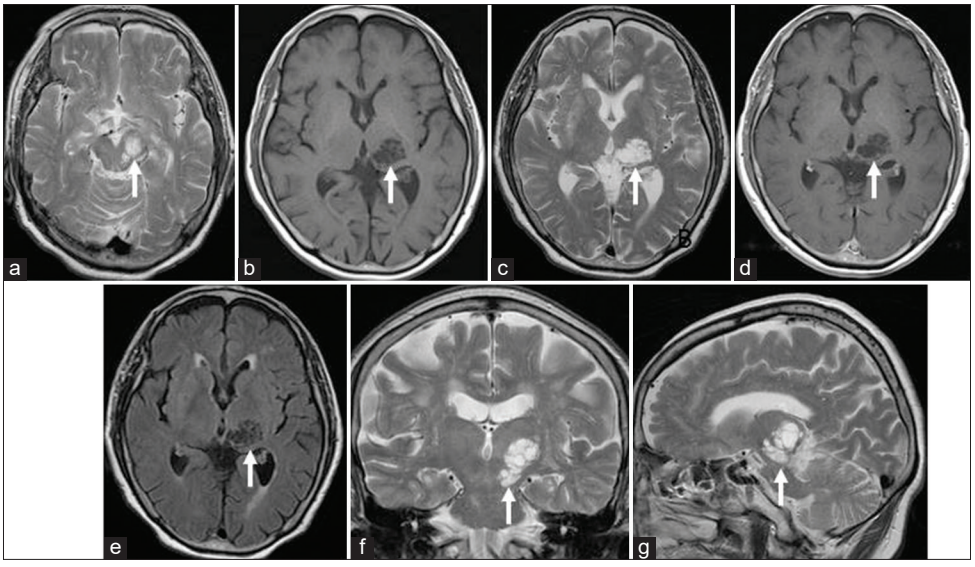

Figure 1: Magnetic resonance image (MRI) of giant perivascular spaces (PVSs) (arrows). (a) Axial T2-weighted image (T2WI) at the level of midbrain. (b-e) Axial images at the level of thalamus. (b) T1WI, (c) T2WI, (d) postgadolinium T1WI, (e) fluid-attenuated inversion recovery image. (f) Coronal T2WI. (g) Sagittal T2WI.

in the territory of the paramedical mesencephalothalamic artery and in the cerebral white matter.[11] Headache is the most common presentation of giant PVSs. Other clinical presentation including oculomotor palsy, trochlear nerve palsy, ataxia, and long tract signs, varies according to their mass effect and location.[1,3,7,11] In addition, obstructive hydrocephalus was seen in about half of these mesencephalothalamic giant PVSs.[1,10]

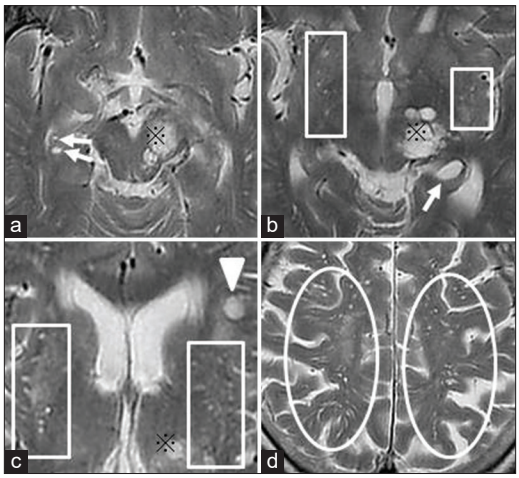

Figure 2: Enlarged PVSs in other locations. ※: Part of the giant PVSs in the mesencephalothalamic region. (a) Right hippocampus (arrows). (b) Bilateral basal ganglia (boxes) and left hippocampus (arrow). (c) Bilateral basal ganglia (boxes) and left frontal operculum (arrowhead). (d) Subcortical white matter (circles).

Pathologies mimicking giant PVSs include pilocytic astrocytomas, gangliogliomas, pleomorphic xanthoastrocytomas, and intraparenchymal schwannoma.[7,8,10,11] The absence of solid part, rim enhancement, surrounding edema, or gliosis and identical intensities of cyst content to cerebral fluid on all MR sequences denied the possibility of neoplasm in our case.

Some rare infectious process results in multiple cyst formation.[11] Actually, measurement of serous antibodies for 12 species of parasite was ordered by a previous attending 3 years ago. All the titers were negative but a single pseudopositivity for strongyloidiasis. The absence of rim enhancement, or surrounding edema, change in signal intensity of cysts' content denied the possibility, too.

Although some giant PVSs must have grown to be symptomatic, no interim change was seen on repeat MRI in 17 cases in which follow-up MRI was studied 4.1 ± 3.7 (mean ± SD) years after the initial scan in Salzmann's series.[11] There were only two literatures which described the growth

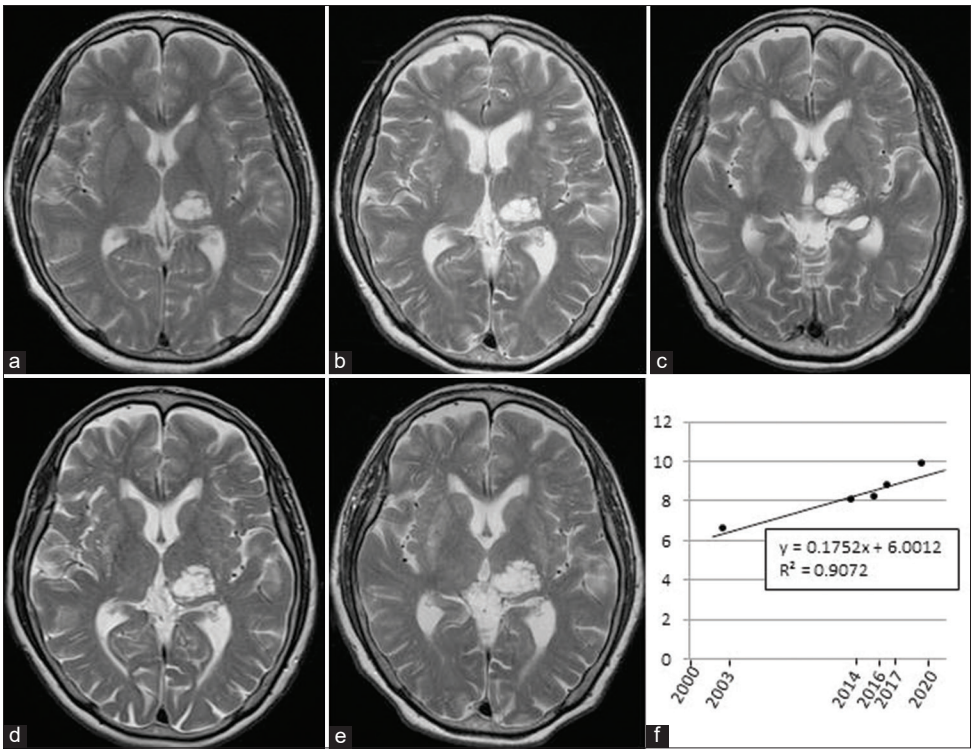

Figure 3: Chronological changes in the volume of the giant perivascular spaces. (a) September 2003: 6.54 ml. (b) May 2014: 8.05 ml. (c) February 2016: 8.19 ml. (d) July 2017: 8.79 ml. (e) June 2020: 9.83 ml. (f) 3D volumetric analysis showing steady and slow growth with gentle slope of linear function: y (ml) = 0.1752x+6.0012, R^2=0.9072.

of the giant PVSs after surgical intervention. Fujimoto *et al*. reported a man with mesencephalic PVSs which had grown very large to manifest oculomotor and trochlear nerve palsies 14 years after the successful third ventriculostomy for the hydrocephalus.[3] Mascalchi *et al*. reported a case in which the mesencephalic PVSs demonstrated new neurological symptoms due to an increase of the number and size of the lesions 4 years after a successful ventriculoperitoneal shunting for symptomatic hydrocephalus.[6] Contrary, Al Abdulsalam *et al*. reported that the size of the lesion decreased following CSF diversion.[1]

The exact etiology of giant PVS enlargement remains unknown, although several possible mechanisms have been suggested in the literature.[1,9,11] Increased CSF pulsation due to ventricular obstruction, an abnormality of arterial wall permeability or obstruction of interstitial fluid drainage pathways has been suggested as contributing factors. Aging and underlying disease condition could be a factor in the pathogenesis of growth of giant PVSs.

This is the first ever reported case showing very gradual enlargement of giant PVSs in a natural course. It has grown steadily by 0.1752 ml/year over 17 years from initial of 6.54 ml. The volume increase rate was 2.68%/year.

Data accumulation of 3-D volumetric analysis may elucidate the true nature of the giant PVSs, which may support to predict the symptomatic growth of incidentally found PVSs.

CONCLUSION

We demonstrated the very slow but steady growth of giant PVSs in the mesencephalothalamic region based on longitudinal MRI follow-up. Data accumulation of 3-D volumetric analysis may elucidate the true nature of the giant PVSs, which may support to predict the symptomatic growth of incidentally found PVSs.

Acknowledgement

The authors would like to thank Haibunsha Kagoshima for English editing.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Al Abdulsalam H, Alatar AA, Elwatidy S. Giant tumefactive perivascular spaces: A case report and literature review. World Neurosurg 2018;112:201-4.
2. Esiri MM, Gay D. Immunological and neuropathological significance of the Virchow-Robin space. J Neurol Sci 1990;100:3-8.
3. Fujimoto K, Kuroda J, Hide T, Hasegawa Y, Yano S, Kuratsu J. Giant tumefactive perivascular spaces that expanded and became symptomatic 14 years after initial surgery. Surg Neurol Int 2012;3:127.
4. Heier LA, Bauer CJ, Schwartz L, Zimmerman RD, Morgello S, Deck MD. Large Virchow-Robin spaces: MR-clinical correlation. AJNR Am J Neuroradiol 1989;10:929-36.
5. Kwee RM, Kwee TC. Virchow-Robin spaces at MR imaging. Radiographics 2007;27:1071-86.
6. Mascalchi M, Salvi F, Godano U, Nistri M, Taiuti R, Tosetti M, *et al*. Expanding lacunae causing triventricular hydrocephalus. Report of two cases. J Neurosurg 1999;91:669-74.
7. McKinney AM. Dilated perivascular spaces. In: Atlas of Normal Imaging Variations of the Brain, Skull, and Craniocervical Vasculature. Vol. 1. Cham, Switzerland: Springer; 2017. p. 315-481.
8. Muzzafar S, Ketonen L, Weinberg JS, Schellingerhout D. Imaging and clinical features of an intra-axial brain stem schwannoma. AJNR Am J Neuroradiol 2010;31:567-9.
9. Poirier J, Barbizet J, Gaston A, Meyrignac C. Thalamic dementia. Expansive lacunae of the thalamo-paramedian mesencephalic area. Hydrocephalus caused by stenosis of the aqueduct of Sylvius. Rev Neurol (Paris) 1983;139:349-58.
10. Rudie JD, Rauschecker AM, Nabavizadeh SA, Mohan S. Neuroimaging of dilated perivascular spaces: From benign and pathologic causes to mimics. J Neuroimaging 2018;28:139-49.
11. Salzman KL, Osborn AG, House P, Jinkins JR, Ditchfield A, Cooper JA, *et al*. Giant tumefactive perivascular spaces. AJNR Am J Neuroradiol 2005;26:298-305.
12. Schley D, Carare-Nnadi R, Please CP, Perry VH, Weller RO. Mechanisms to explain the reverse perivascular transport of solutes out of the brain. J Theor Biol 2006;238:962-74.
13. Weller RO, Djuanda E, Yow HY, Carare RO. Lymphatic drainage of the brain and the pathophysiology of neurological disease. Acta Neuropathol 2009;117:1-14.

How to cite this article: Okada T, Makimoto K, Itoh K, Moinuddin FM, Yoshimoto K, Arita K. The first 3-D volumetric analysis of mesencephalothalamic giant perivascular spaces showing steady and slow growth over 17 years. Surg Neurol Int 2020;11:300.