

Two patients with isolated dysarthria caused by cerebellar infarction

Case Report

Katsuhiko Ogawa*, Yutaka Suzuki, Minoru Oishi, Satoshi Kamei

Division of Neurology, Department of Medicine, Nihon University, School of Medicine, Tokyo. 173-8610, Japan

Received 20 April 2012; Accepted 3 May 2013

Abstract: Introduction: The paravermal zone of the right rostral cerebellar hemisphere is an important area for speech function. Recent functional MRI (fMRI) studies reported the significance of lobulus simplex within the rostral cerebellum for speech function. Here, we assess the responsible lesion for dysarthria within the rostral cerebellum. Patients and methods: In order to evaluate the lesion in the rostral cerebellum responsible for cerebellar dysarthria, we compared the locations of infarcts from 4 reported patients with isolated dysarthria caused by cerebellar infarction as well as an additional newly encountered patient in whom cerebellar infarction caused isolated dysarthria. Results: The paravermal zone of the rostral cerebellar hemisphere was involved in all 5 patients. The lesions were located in the lobulus quadrangularis and lobulus simplex in 3 of the patients, and the lobulus simplex and lobulus semilunaris superior in 2 of the patients. The infarcts in 4 of the patients were located on the right side. Conclusion: The lesions in 4 of the patients included the right paravermal zone of the rostral cerebellar hemisphere, which is regarded as an important area for speech function. The lobulus simplex was most highly involved in all 5 patients. We present the importance of lobulus simplex for speech function, as in the fMRI data of previous reports, based on the localization of cerebellar infarcts in patients with isolated dysarthria.

Keywords: Cerebellar infarction • Isolated dysarthria • Lobulus simplex

© Versita Sp. z o.o

1. Introduction

Dysarthria is a frequent neurological consequence of cerebellar diseases [1]. The cerebellum is supplied by 3 arteries, the superior cerebellar artery (SCA), the anterior inferior cerebellar artery (AICA), and the posterior inferior cerebellar artery (PICA). Dysarthria is most common in infarctions within the territory of the SCA among the 3 arteries [2–7]. In the territory of the SCA, the involvement of the paravermal zone in the right rostral cerebellar hemisphere, supplied by the rostral trunk of the SCA [8], has been indicated in the occurrence of dysarthria [9,10]. Recent functional MRI (fMRI) data emphasized the right cerebellar hemisphere as well as the dorsolateral and medial areas of the left (language-dominant) frontal lobe for speech function [11]. The localization of speech function in the rostral

cerebellum has been reported in fMRI studies [12,13]. These fMRI studies showed that speech production relied on the paravermal lobulus simplex in the rostral cerebellum [12,13]. Previously, we reported a patient with isolated dysarthria caused by cerebellar infarction. We hypothesized that the lobulus simplex might be the responsible lesion in the cerebellum for dysarthria [14], and emphasized the paravermal zone of the rostral cerebellar hemisphere for cerebellar dysarthria [15]. In this report, we have included an additional patient who showed isolated dysarthria caused by cerebellar infarction. We studied the responsible lesion for dysarthria in the cerebellum, based on the localization of the lesions in patients with isolated dysarthria caused by cerebellar infarction [2, 14–17], and the recently reported fMRI data on speech function in the cerebellum [12, 13].

* E-mail: ogawa.katsuhiko@nihon-u.ac.jp

2. Patients and methods

Two patients were reviewed who showed isolated dysarthria caused by cerebellar infarction (Patient 1 was previously reported [14,15]). In addition, 3 other patients with isolated dysarthria caused by cerebellar infarction were previously reported (Table 1) [2,16,17]. The radiological findings (MRI) of our 2 patients (One patient was previously reported [14,15].) and the other 3 previously reported patients were analyzed (Table 1). The locations of infarcts of these 5 patients were compared to consider the region mainly related to speech function within the cerebellum.

3. Results

3.1. Patient 1 [14, 15]

A right-handed 69-year-old man was admitted to the cardiosurgery service of our hospital due to exercise-induced pain in the right upper extremity. He was diagnosed as having arteriosclerotic obliteration (ASO) in the right upper extremity. Pain in the right upper extremity improved by intravenous administration of prostaglandin. Two months after the onset of ASO, the patient again complained of pain in the right upper extremity. He was admitted to our hospital and underwent emergent angiography of the right extremity, and was discharged the same day. However, speech disturbance developed at night following the discharge.

He was admitted to the neurology ward the next day due to the persistent speech disturbance. Computed tomography (CT) of the brain demonstrated an infarct in the right cerebellar hemisphere. On admission, the rhythm of his speech was moderately poor. The initial words of his speech occasionally became explosive. The speed of his speech was moderately slow. Scanning speech was also present. Finger-to-nose test and heel-knee-shin test were normal on both sides. Truncal and gait ataxia was not present. General and other neurological findings were normal, including the cranial nerves, muscle strength and tone, and sensation. There were no involuntary movements, orthostatic hypotension or vesicorectal dysfunction. Findings on blood cell counts and chemistry, urine, electrocardiogram, and cardioechogram were normal. Cranial T2-weighted MRI demonstrated an infarct at the paravermal zone in the rostral cerebellar hemisphere on the right side (Figure 1). The localization of the lesion was consistent with the lobulus quadrangularis and lobulus simplex (Figure 1). No lesions were detected in the supratentorial regions or the brainstem. Dysarthria improved gradually and disappeared 5 days after hospitalization.

3.2. Patient 2

A right-handed 81-year-old man with history of hyperlipidemia developed sudden onset of headache and dysarthria and was admitted to our hospital. On admission, scanning speech was moderately present. The rhythm of his speech was moderately poor. The finger-to-nose test and heel-knee-shin test were normal on both sides.

Table 1. Cerebellar infarction with isolated dysarthria

Author	age (year)/sex	dominant hand	side of lesion	paravermal zone of the rostral cerebellar hemisphere	region of infarct localization of infarct in the cerebellar hemisphere			prognosis
					lobulus quadrangularis	lobulus simplex	lobulus semilunaris superior	
Kuwabara et al.(1992) [2]	58/male	unknown	right	involved	+	+	–	unknown
Amarenco et al.(1991) [17]	69/male	unknown	left	involved	–	+	+	unknown
Gironell et al.(1996) [16]	78/male	right	right	involved	+	+	–	good
Patient 1(2004, 2010) [14, 15]	69/male	right	right	involved	+	+	–	good
Patient 2	81/male	right	right	involved	–	+	+	good

Truncal and gait ataxia was not present. Deep tendon reflexes of the four extremities were slightly decreased. General and other neurological findings were normal, including the cranial nerves, muscle strength and tone, and sensation. There were no involuntary movements,

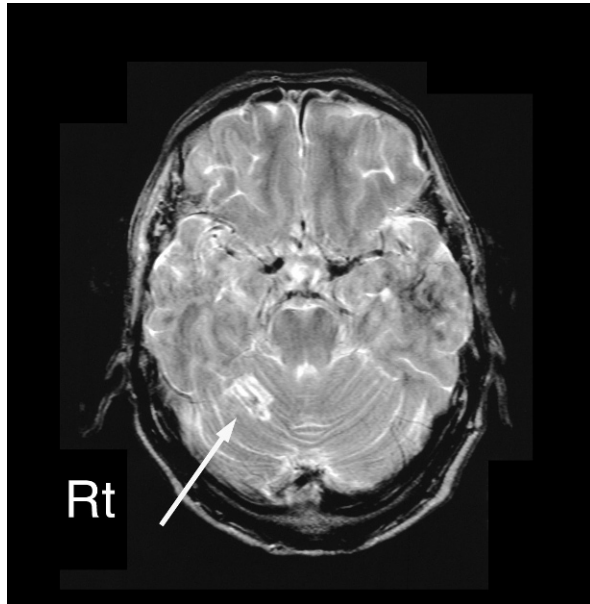


Figure 1. T2-weighted cranial MRI (TR 3600 msec / TE 96 msec). An infarct was located in the lobulus quadrangularis and lobulus simplex on the right side. Paravermal zone was involved in the infarct.

orthostatic hypotension or vesicorectal dysfunction. Findings on blood tests showed a mild increase of total cholesterol. Findings on urine, electrocardiogram, and cardioechogram were normal. Cranial CT demonstrated an infarct at the cerebellar hemisphere on the right side. Cranial T1-weighted MRI showed an infarct at the paravermal zone in the rostral cerebellar hemisphere on the right side (Figure 2). The localization of the lesion was consistent with the lobulus simplex (Figure 2A) and lobulus semilunaris superior (Figure 2B). No lesions were detected in the supratentorial region or the brainstem. Dysarthria improved gradually and disappeared after 2 weeks of hospitalization.

4. Discussion

The cerebellum is supplied by the SCA, AICA, and PICA. Rostral cerebellum is supplied by the SCA. The SCA arises as a single trunk from the basilar artery and then bifurcates into the rostral trunk and the caudal trunk [8]. The rostral trunk supplies the vermis and the paravermian area of the rostral cerebellum [8]. The common clinical findings of SCA infarcts are dysarthria and limb ataxia [2–6]. Vertigo, headache, and nystagmus are less common in SCA infarcts than in PICA infarcts [4]. In the rostral cerebellum, dysarthria is mainly caused

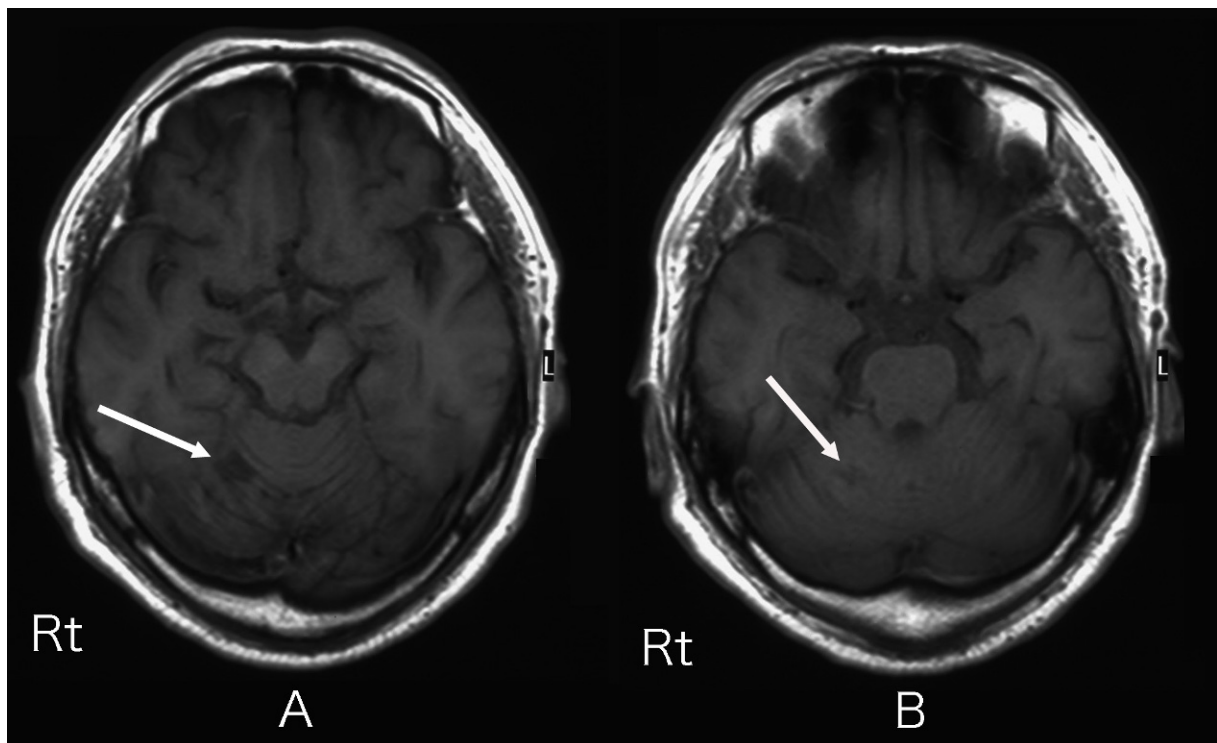


Figure 2. T1-weighted cranial MRI (TR 631 msec / TE 13 msec). An infarct was located in the lobulus simplex (A) and lobulus semilunaris superior (B) of the upper cerebellar hemisphere on the right side. Paravermal zone was involved in the infarct.

by the impairment of the right paravermal zone of the rostral cerebellar hemisphere [9,10] where the rostral trunk supplies in the territory of SCA [8]. Urban PP et al. reported that lesions in the upper paravermal zone of the right cerebellar hemisphere, the site of coordination of articulatory movements of the tongue and orofacial muscles, might lead to the development of dysarthria, unrelated to brainstem infarction [9]. The infarcts of the 2 patients we report were also located in paravermal zone of the rostral cerebellar hemisphere on the right side.

Cerebellar dysarthria is called ataxic speech. Ataxic speech is characterized by slurred speech, explosive staccato scanning vocalization, and wavering modulation [18]. The symptoms of dysarthria in our 2 patients were consistent with the characteristics of ataxic speech.

Five patients with pure dysarthria due to cerebellar infarction, including Patient 1, have been reported previously [2,14–17]. The clinical findings of the 5 patients with isolated dysarthria caused by cerebellar infarction are listed in the table [2,14–17] (Table 1). The infarcts in the 5 patients involved the paravermal zone of the rostral cerebellar hemisphere supplied by the rostral trunk of the SCA. Out of these 5 patients [2,14–17], infarcts in the 4 patients were located on the right side [2,14–16]. This result was consistent with the previous reports that emphasized the paravermal zone in the rostral cerebellar hemisphere on the right side for speech function [9, 10]. The infarcts in the reported patient of Amarenco et al. [17] and Patient 2 were localized in the lobulus simplex and lobulus semilunaris superior. The infarcts in the other 3 patients [2,14–16] were anatomically localized in the lobulus quadrangularis and lobulus simplex. Localization of speech function in the upper cerebellum has been studied in previous reports [12,13]. Frings M et al. reported that speech production relied on the paravermal zone of the bilateral lobulus simplex, whereas verb generation was associated with the lateral hemispherical part of the lobulus simplex, based on their fMRI study [13]. Dresel C et al. reported a fMRI study of whistling [12]. They showed a significant correlation between the whistle amplitude and activation in the right

premotor cortex and lobulus simplex in the right rostral paravermal cerebellar hemisphere [12]. In our study, the lobulus simplex was the most impaired lesion in all of the 5 patients (Table 1). We therefore considered that the lobulus simplex was mainly related to speech function. This result was consistent with the fMRI data of the previous 2 reports that emphasized the lobulus simplex for speech function [12, 13].

Ackermann et al. emphasized the right cerebellar hemisphere for speech function, based on the fMRI findings [11,19]. They reported that the right cerebellar hemisphere connected with the lower precentral gyrus in the left frontal lobe [11,19]. Dominant hand was described in our 2 patients and a reported patient of Gironel et al [14–16]. The dominant hand was right in the 3 patients (Table 1). We considered that the right cerebellar hemisphere which connected with the left (language-dominant) frontal lobe dominated for speech function in the 3 patients.

The majority of patients (80% to 95%) with cerebellar infarction are reported to have a benign clinical course [20], while cerebellar signs and symptoms in the patients with spinocerebellar degeneration are progressive. Prognosis of dysarthria was shown in our 2 patients and a separate patient reported by Gironel et al [14–16]. These 3 patients also showed a good recovery and their dysarthria improved completely.

5. Conclusion

The paravermal zone in the rostral cerebellar hemisphere was involved in all of the patients reviewed with isolated dysarthria (Table 1) and was found to be more impaired on the right side. This result was consistent with the previous reports indicating the right paravermal zone in the rostral cerebellar hemisphere for speech function [12,13]. We showed that the lobulus simplex was the most impaired lesion and considered that the lobulus simplex was mainly related to speech function in the cerebellum, as did previous reports that showed the importance of the lobulus simplex for speech function [12,13].

References

- [1] Urban P.P., Rolke R., Wicht S., Keilmann A., Stoeter P., Hopf H.C., et al., Left-hemispheric dominance for articulation: a prospective study on acute ischaemic dysarthria at different localizations. *Brain* 2006 ; 129 : 767-777
- [2] Kuwahara S., Hirayama K., Kojima S., Kawamura S., Clinical features of infarction in the territory of the superior cerebellar artery. *Jpn J Stroke* 1992 ; 14 : 159-165 (in Japanese with English abstr)
- [3] Terao S., Sobue G., Izumi M., Miura N., Mitsuma T., Cerebellar infarction in the territory of the superior cerebellar artery, presenting a predominant cerebellar symptom – with special reference to its pathophysiology –. *Clin Neurol* 1995 ; 35 : 256-261
- [4] Chaves C.J., Caplan L.R., Chung C.S., Amarenco P., Cerebellar infarcts. *Current Neurology* 1994;14: 143-177

- [5] Erdemoglu A.K., Duman T., Superior cerebellar artery territory stroke. *Acta Neurol Scand* 1998 ; 98: 283-287
- [6] Barth A., Bogousslavsky J., Regli F., The clinical and topographic spectrum of cerebellar infarcts : a clinical-magnetic resonance imaging correlation study. *Ann Neurol* 1993 ; 33 : 451-456
- [7] Cano L.M., Cardona P., Quesada H., Mora P., Rubio F., Cerebellar infarction: prognosis and complications of vascular territories. *Neurologia* 2012 ; 27 : 330-335
- [8] Rhoton A.L. Jr., The cerebellar arteries. *Neurosurgery* 2000 ; 47 (3 Suppl) : S29-68
- [9] Urban P.P., Marx J., Hunsche S., Gawehn J., Vucurevic G., Wicht S., et al., Cerebellar speech representation. Lesion topography in dysarthria as derived from cerebellar ischemia and functional magnetic resonance imaging. *Arch Neurol* 2003 ; 60 : 965-972
- [10] Lechtenberg R., Gilman S., Speech disorders in cerebellar disease. *Ann Neurol* 1978 ; 3 : 285-290
- [11] Ackermann H., Mathiak K., Riecker A., The contribution of the speech production and speech perception: clinical and functional imaging data. *Cerebellum* 2007 ; 6 : 202-213
- [12] Dresel C., Castrop F., Haslinger B., Wohlschlaeger A.M., Hennenlotter A., Ceballos-Baumann A.O., The functional neuroanatomy of coordinated orofacial movements: sparse sampling fMRI of whistling. *Neuroimage* 2005 ; 28 : 588-597
- [13] Frings M., Dimitrova A., Schorn C.F., Elles H.G., Hein-Kropp C., Gizewski E.R., et al., Cerebellar involvement in verb generation: an fMRI study. *Neurosci Lett* 2006 ; 409 : 19-23
- [14] Ogawa K., Suzuki Y., Kamei S., Mizutani T., A case of cerebellar infarction with pure dysarthria. *Clin Neurol* 2004 ; 44 : 111-113 (in Japanese with English abstr)
- [15] Ogawa K., Yoshihashi H., Suzuki Y., Kamei S., Mizutani T., Clinical study of the responsible lesion for dysarthria in the cerebellum. *Inter Med* 2010 ; 49 : 861-864
- [16] Gironell A., Arboix A., Marti-Vilalta J.L., Isolated dysarthria caused by a right paravermal infarction. *J Neurol Neurosurg Psychiatry* 1996 ; 61 : 205-206
- [17] Amarenco P., Chevrie-Muller C., Rouillet E., Bousser M.G., Paravermal infarct and isolated cerebellar dysarthria. *Ann Neurol* 1991 ; 30 : 211-213
- [18] Chevrie-Muller C., Guidet C., RÔle de cervelet dans le contrôle de la hauteur de la voix et de ces modulations. In : Boë L.J., Descout R., Guérin B. (Eds.) *Larynx et Paroles*. Bull Instit Phonet Grenoble 1979 ; 8 : 3-26
- [19] Ackermann H., Cerebellar contributions to speech production and speech perception: psycholinguistic and neurobiological perspectives. *Trends in Neurosciences* 2008 ; 31 : 265-272
- [20] Fujimoto M., Atsumi T., Cerebellar infarction. *Shinkeinaika* 1997 ; 47 : 468-474 (in Japanese with English abstr)