Case Report

Mutism resulting from heterochronic bilateral cerebellar hemorrhages – A case report

Masahito Katsuki1, Ayumi Narisawa1, Hiroshi Karibe1, Motonobu Kameyama1, Teiji Tominaga2
Departments of Neurosurgery, 1Sendai City Hospital, 1-1-1, Asuto-Nagamachi, Taihaku-ku, 2Tohoku University Graduate School of Medicine, 1-1 Seiryo, Aobaku, Sendai, Japan.
E-mail: Masahito Katsuki – ktk1122nigt@gmail.com; Ayumi Narisawa – ayunarisawa@gmail.com; *Hiroshi Karibe – karibe@nsg.med.tohoku.ac.jp; Motonobu Kameyama – motokame@qk9.so-net.ne.jp; Teiji Tominaga – tomi@nsg.med.tohoku.ac.jp

INTRODUCTION

Cerebellar mutism (CM) is a neurological condition characterized by lack of speech despite reading and writing with intact comprehension after cerebellar injury[6,16,18] lasting for a few months on average.6,16,18 CM rarely occurs in children after surgery for posterior fossa midline tumors[16] such as medulloblastoma.17 CM also occurs following trauma,9 stroke,3,5,8,10,11,13,15,16 and infections.14 Interruptions of the bilateral dentatothalamocortical pathways (DTC) may be the principal cause of CM.1,18 CM can result from injuries anywhere along the DTC,9 However, the anatomical substrate of CM is not fully understood. It has also been under debate whether bilateral, unilateral, or dominant side injury of the DTC is the cause of CM. The previous CM patients generally underwent neurosurgical manipulation to the cerebellum, resulting in difficulty to indicate the responsible region precisely.
We report a CM patient with both a recent right cerebellar hemorrhage and an older left one. The head computed tomography (CT) or magnetic resonance imaging (MRI) showed injuries to the bilateral dentate nuclei. Previous reports are scarce on CM resulting from injuries of the bilateral dentate nuclei. This report can contribute to the elucidation of the mechanism of CM and support the hypothesis that bilateral interruption of the DTC is the principal cause of CM.

**CASE REPORT**

An 87-year-old right-handed female patient presented to our emergency room with depression of alertness after sudden vomiting. She had suffered from the left cerebellar hemorrhage 20 years ago and had been taking antihypertensive medications. Her blood pressure was 235/135 mmHg on admission. She had mild dysmetria and did not speak any words although she could obey commands, with the Glasgow coma scale score of 11 (E4V1M6). The head CT showed a high-density area in the right cerebellar hemisphere, presenting 9 mL of a right cerebellar hematoma. It also showed a low-density area in the left cerebellar hemisphere, presenting an older left cerebellar hemorrhage. The head MRI showed perifocal edema surrounding the right cerebellar hematoma. Both the thalamus and the supplementary motor area were bilaterally intact on both CT and MRI [Figure 1].

On the following day, her neurological findings did not change and rehabilitation began. The CT did not show hematoma expansion. On the 3rd day, she was mute but cognitively alert, although she could communicate by nodding, shaking her head, or facial expression enabled communication.

She could not walk independently and was transferred to a rehabilitation hospital 31 days after the onset of the right cerebellar hemorrhage. Her mutism improved a bit and a few short words (e.g. yes and no) were uttered. Four months after the onset, she could walk with a little care. However, her mutism did not change compared to when she was transferred to the rehabilitation hospital. The CT and MRI did not show any additional findings rather than the injuries of the bilateral dentate nuclei. She was diagnosed as CM caused by heterochronic bilateral cerebellar hemorrhages.

**DISCUSSION**

In general, mutism is defined as the inability to speak despite cognitively alert patient. It can occur due to lesions in several parts of the brain, such as Broca’s area, the supplementary motor area, thalamus, mesencephalic reticular formation regions, and bilateral hemispheric lesions. CM is one of the types of transient mutism resulting from interruptions of the bilateral DTC pathways, in general. Posterior fossa tumor surgery is the most common cause of this syndrome, as symptoms usually last for a few months. Surgical excision of the posterior fossa tumor causes CM, occurring in 27.7% and its duration is limited from a few weeks until 120 days. In the present case, surgical treatment was not performed, and the CM lasted for more than 4 months. It seemed different from the typical CM in its clinical characteristics. Thus, the mechanism of CM is discussed below focusing on the bilateral DTC including dentate nuclei.

The anatomical substrate of CM is not fully understood. Vermis, brainstem, and anywhere along the DTC are reported as a candidate for responsible regions of CM after posterior fossa surgery. The vermis has neural connections with the deep cerebellar nucleus and is involved in the coordination of laryngeal and respiratory function. The inferior vermis seems to be associated with speech initiation, suggesting that splitting of the vermis was related to CM. However, either the cerebellar hemisphere or the brain stem could be manipulated to some extent intraoperatively. Therefore, it seems difficult to attribute CM only to the vermis.

CM seems to be also related to brainstem involvement of the tumor. The DTC connects the dentate nucleus to the contralateral cerebral cortex. The nerve passes the decussation of the superior cerebellar peduncle, the red nucleus in the midbrain, and the ventrolateral anterior nucleus of the thalamus [Figure 2a]. Therefore, direct damage or postoperative edema in the brainstem would interrupt the DTC bilaterally, causing CM [Figure 2b].
Katsuki, et al.: Mutism after heterochronic bilateral cerebellar hemorrhages

There have been six previous case reports of CM after cerebellar hemorrhage [Table 1].

Although the sites of cerebellar hemorrhage were unilateral in five of six reports, all of the hematomas were so massive that they invaded beyond the midline structures, requiring surgical interventions in most cases. Either the massive hematoma and/or the surgical intervention would injure both the unilateral dentate nucleus and the brainstem or the contralateral superior cerebellar peduncle. Therefore, the bilateral DTC would have been injured even in cases with unilateral hemorrhage [Figure 2c].

In the present case, injuries were limited in the bilateral dentate nuclei due to the recent right cerebellar hemorrhage and the older left cerebellar hemorrhage without surgical treatment. Besides, the CT and MRI did not show any other lesions along the DTC. In previous reports on CM, due to surgical intervention or massive hemorrhage, it has been difficult to indicate the responsible region for CM precisely. The present case seems simple and can support the theory that bilateral interruptions of the DTC are the cause of CM [Figure 2d].

Some cases of CM resulting from injuries of bilateral dentate nuclei have been reported. Kubota et al. reported CM due to rotavirus-associated acute cerebellitis.[12] The MRI showed the signal changes in the bilateral dentate nuclei. Guidetti and Fraioli performed stereotactic manipulation to the bilateral dentate nuclei as a treatment for dyskinesia. They observed mutism in two of their 88 patients 1–3 months after the surgery.[7] This is the only report of CM due to direct injuries to the bilateral dentate nuclei. Therefore, our report is the first report on CM due to lesions of the bilateral dentate nuclei without surgical manipulation.

CONCLUSION

We reported the patient with CM due to the recent right cerebellar hemorrhage and the older left one. Both CT and MRI did not show any lesions other than the bilateral dentate

---

Table 1: Case series of cerebellar mutism due to cerebellar hemorrhage.[3,5,8,10,13,15]

| Author (Year) | Age/Sex | Pathology | Site     | Medial or contralateral invasion | Treatment          |
|---------------|---------|-----------|----------|----------------------------------|--------------------|
| Coplin (1997) | 47/Male | Unknown   | Supravermis | +                                | Hematoma removal   |
| Kawai (2001)  | 64/Male | HT        | R        | +                                | Hematoma removal   |
| Idiaquez (2011)| 20/Male | AVM       | L        | +                                | Hematoma removal   |
| De Smet (2012)| 60/Male | HT        | L        | +                                | Hematoma removal   |
| Marién (2013) | 71/Male | HT        | R        | +                                | Drainage           |
| Lee (2017)    | 20/Female | AVM     | L        | +                                | Hematoma removal   |
| Our case (2018)| 87/Female | HT     | R, older L | -                                | Medical treatment  |

AVM: Arteriovenous malformation, HT: Hypertension, L: Left, R: Right
nuclei. This report can support the hypothesis that bilateral interruptions of the DTC are the cause of CM.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Acknowledgements

Conception and design: Hiroshi Karibe. Drafting the article: all authors. Supervision: Hiroshi Karibe, Teiji Tominaga.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Ackermann H, Mathiak K, Riecker A. The contribution of the cerebellum to speech production and speech perception: Clinical and functional imaging data. Cerebellum 2007; 6:202-13.
2. Catsman-Berrevoets CE, Aarsen FK. The spectrum of neurobehavioural deficits in the posterior fossa syndrome in children after cerebellar tumour surgery. Cortex 2010; 46:933-46.
3. Coplin WM, Kim DK, Kliot M, Bird TD. Mutism in an adult following hypertensive cerebellar hemorrhage: Nosological discussion and illustrative case. Brain Lang 1997;59:473-93.
4. Dailey AT, McKhann GM 2nd, Berger MS. The pathophysiology of oral pharyngeal apraxia and mutism following posterior fossa tumor resection in children. J Neurosurg 1995;83:467-75.
5. De Smet HJ, Mariën P. Posterior fossa syndrome in an adult patient following surgical evacuation of an intracerebellar hematoma. Cerebellum 2012;11:587-92.
6. Gudrunardottir T, Sehested A, Juhler M, Schmiegelow K, Cerebellar mutism: Review of the literature. Childs Nerv Syst 2011;27:355-63.
7. Guidetti B, Fraioli B. Neurosurgical treatment of spasticity and dyskinesias. Acta Neurochir (Wien) 1977;24:27-39.
8. Idiaquez J, Fadic R, Mathias CJ. Transient orthostatic hypertension after partial cerebellar resection. Clin Auton Res 2011;21:57-9.
9. Kariyattil R, Rahim MI, Muthukuttiparambil U. Cerebellar mutism following closed head injury in a child. Sultan Qaboos Univ Med J 2015;15:133-5.
10. Kawai H, Ohta F, Matsumoto Y, Yamamoto Y. An adult case of cerebellar mutism after removal of cerebellar hematoma. No To Shinkei 2001;53:475-9.
11. Koh S, Turkel SB, Baram TZ. Cerebellar mutism in children: Report of six cases and potential mechanisms. Pediatr Neurol 1997;16:218-9.
12. Kubota T, Suzuki T, Kitase Y, Kidokoro H, Miyajima Y, Ogawa A, et al. Chronological diffusion-weighted imaging changes and mutism in the course of rotavirus-associated acute cerebellitis/cerebellopathy concurrent with encephalitis/encephalopathy. Brain Dev 2011;33:21-7.
13. Lee S, Na YH, Moon HI, Tae WS, Pyun SB. Neuroanatomical mechanism of cerebellar mutism after stroke. Ann Rehabil Med 2017;41:1076-81.
14. Makarenko S, Singh N, McDonald PJ. Non-surgical transient cerebellar mutism-case report and systematic review. Childs Nerv Syst 2018;34:535-40.
15. Mariën P, Verslegers L, Moens M, Dua G, Herregods P, Verhoeven J, et al. Posterior fossa syndrome after cerebellar stroke. Cerebellum 2013;12:686-91.
16. Nishikawa M, Komiyama M, Sakamoto H, Yasui T, Nakajima H. Cerebellar mutism after basilar artery occlusion case report. Neurol Med Chir (Tokyo) 1998;38:569-73.
17. Reed-Berendt R, Phillips B, Picton S, Chumas P, Warren D, Livingston JH, et al. Cause and outcome of cerebellar mutism: Evidence from a systematic review. Childs Nerv Syst 2014;30:375-85.
18. Tamburrini G, Frassanito P, Chieffo D, Massimi L, Caldarelli M, Di Rocco C, et al. Cerebellar mutism. Childs Nerv Syst 2015;31:1841-51.
19. van Baarsen KM, Grotenhuis JA. The anatomical substrate of cerebellar mutism. Med Hypotheses 2014;82:774-80.

How to cite this article: Katsuki M, Narisawa A, Karibe H, Kameyama M, Tominaga T. Mutism resulting from heterochronic bilateral cerebellar hemorrhages – A case report. Surg Neurol Int 2019;10:122.