Radiology Case Reports 19 (2024) 4610–4613

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journal homepage: www.elsevier.com/locate/radcr

Case Report

Recurrent hiccups associated with ipsilateral intracerebral hemorrhage and chronic subdural hematoma with immediate resolutions after evacuations ☆,☆☆,★

Kasumi Inami, MD, Satoshi Tsutsumi, MD, Hana Asagiri, MD, Motoki Yamataka, MD, Natsuki Sugiyama, MD, Hideaki Ueno, MD, Hisato Ishii, MD*

Department of Neurological Surgery, Juntendo University Urayasu Hospital, Urayasu, Chiba, Japan

ARTICLE INFO

Article history:
Received 23 May 2024
Revised 14 July 2024
Accepted 15 July 2024

Keywords:
Central hiccup
Intracerebral hemorrhage
Chronic subdural hematoma
Intracranial pressure

ABSTRACT

A 74-year-old man presented with persistent hiccups and headache persisting for 2 days. An anticoagulant was administered for his coronary heart disease. Cranial computed tomography (CT) revealed an intracerebral hemorrhage (ICH) located in the right occipital lobe, without any abnormal findings around the brainstem. The patient underwent endoscopic hematoma evacuation via a burr hole, resulting in immediate resolution of hiccups. Following an uneventful postoperative course, the patient experienced recurrent hiccups on the 47th day postsurgery. A subsequent CT scan taken on the 50th day revealed a compressive chronic subdural hematoma (CSDH) situated in the right frontoparietal convexity. The patient underwent burr-hole irrigation, leading to prompt cessation of the hiccups. Persistent hiccup should be recognized as potential manifestation of supratentorial lesions, including ICH or CSDH. Surgical evacuation of such lesions can rapidly alleviate hiccups associated with these pathologies.

Introduction

Hiccups manifest as sudden, involuntary contractions of the diaphragmatic and intercostal muscles, promptly succeeded by laryngeal closure [1,2]. It is widely postulated that a reflex arc involving peripheral phrenic, vagal, and sympathetic pathways, alongside central midbrain modulation, underlies the genesis of hiccups. Cardiovascular hiccups, a peripheral type of hiccup, often stem from myocardial infarction or ischemia. Whereas, hiccups primarily developing from the central nervous system, the central hiccups, are predominantly

☆ Competing Interests: The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

☆☆ Acknowledgements: The authors have no conflicts of interest to declare regarding the materials or methods in this study or the findings specified in this paper.

★ Disclosure of Funding: None.

* Corresponding author.
E-mail address: shotaro@juntendo-urayasu.jp (S. Tsutsumi).

https://doi.org/10.1016/j.radcr.2024.07.081

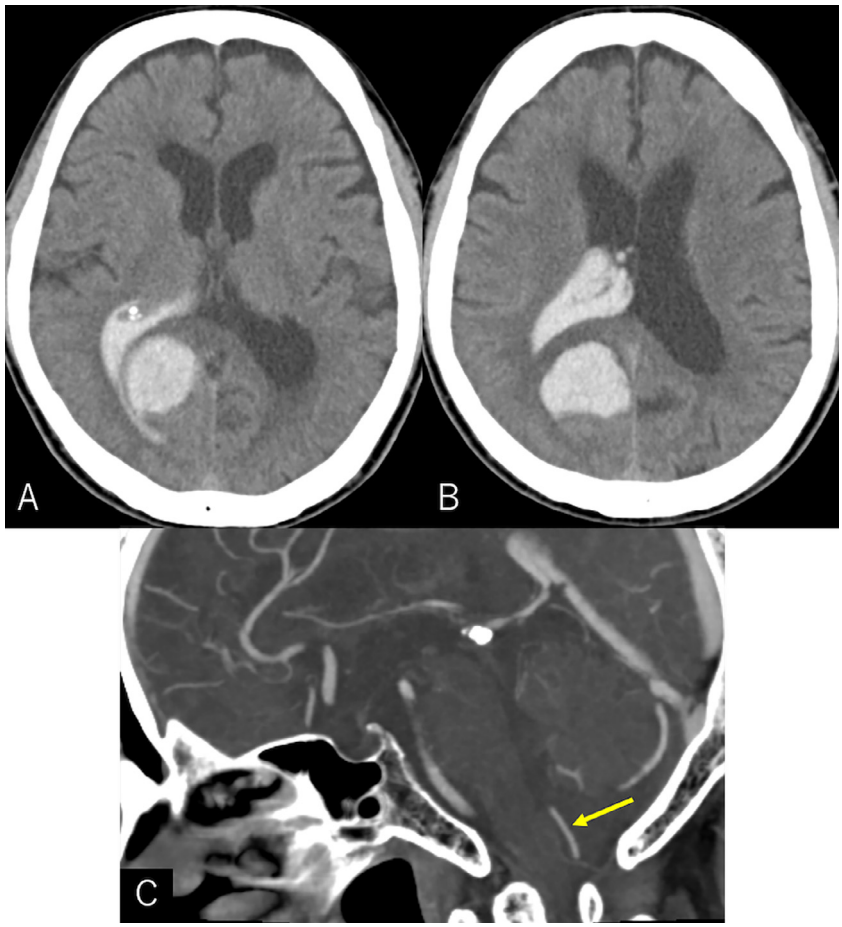

Fig. 1 – Noncontrast axial computed tomography scans at the level of the foramen of Monro (*A*) and body of the lateral ventricle (*B*), performed at the initial presentation, depict a 33 × 24 × 43 mm intracerebral hemorrhage in the right occipital lobe, accompanied by ventricular perforation. (*C*) Contrast-enhanced sagittal computed tomography scan reveals no abnormal findings around the brainstem (*arrow*).

associated with various lesions affecting the dorsal medulla oblongata [1,3–7]. As rare instances of the central hiccups, supratentorial conditions such as tumors, infarctions, abscesses, and hydrocephalus have been documented to cause hiccups [4,8–14].

In this report, we present an intriguing case of persistent hiccups caused by spontaneous intracerebral hemorrhage (ICH) and subsequently complicated by ipsilateral chronic subdural hematoma (CSDH), which notably resolved immediately after evacuation.

Case report

A 74-year-old man presented with persistent hiccups and headaches over 2 days. He had a medical history for coronary heart disease and had been administered anticoagulant therapy (clopidogrel 75 mg daily). Upon examination, the patient was well oriented but demonstrated left homonymous hemianopsia. He was not aware of chest symptoms with normal electrocardiographic findings. Cranial computed tomography (CT) scans revealed an ICH measuring 33 × 24 × 43 mm in maximal diameter, located in the right occipital lobe, and accompanied by ventricular perforation. Further evaluation with contrast-enhanced CT ruled out any vascular lesions or tumors. There were no abnormal findings in or around the brainstem (Fig. 1). The patient subsequently underwent endoscopic hematoma evacuation via burr hole that resulted in an immediate resolution of hiccups and headache. A follow-up CT performed on the 40th postoperative day indicated thin subdural fluid accumulation in the right frontal convexity (Fig. 2). The patient remained asymptomatic at this time. However, on the 47th day postoperatively, he experienced recurrence of persistent hiccups that prompted presentation on the 50th day. At the examination, the patient was well oriented without identifiable motor weakness in the upper or lower extremities. Also, he denied headaches or nausea. However, CT performed on the same day revealed compressive CSDH in the right frontoparietal convexity. It measured 23 mm in maximum thickness, predominantly appearing isodense, and accompanied by fluid-fluid level (Fig. 3). An emergent burr-hole irrigation was performed, leading to an immediate cessation of the hiccups. CT on the following day demonstrated successful removal of the CSDH (Fig. 4). Currently, the patient has been monitored for 6 months without recurrence of hiccups.

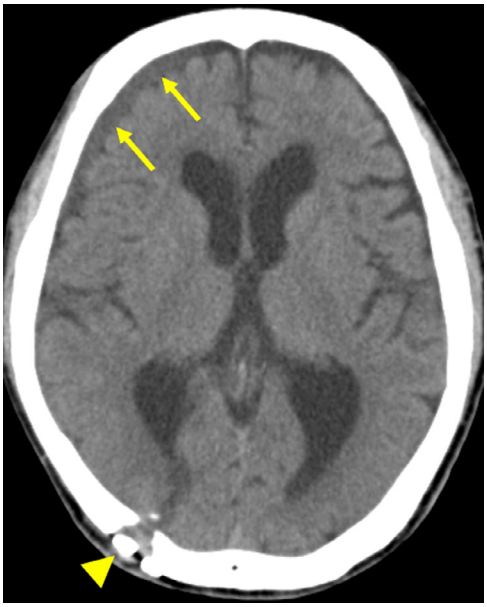

Fig. 2 – Noncontrast axial computed tomography scan on the 40th postoperative day demonstrates complete resolution of intracerebral and intraventricular hemorrhages. In addition, thin subdural fluid accumulation is evident in the right frontal convexity (*arrows*). *Arrowhead*: burr-hole.

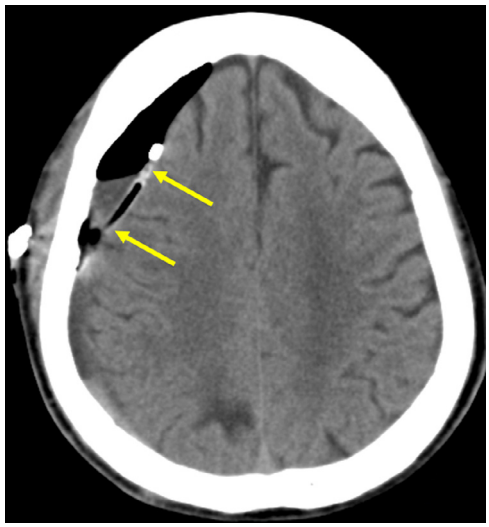

Fig. 4 – Noncontrast axial computed tomography scan taken on the day following burr-hole irrigation displays satisfactory removal of the subdural hematoma. *Arrows*: subdurally placed drain during surgery.

Discussion

In the present case, 2 consecutive episodes of persistent hiccups occurring in association with a supratentorial ICH and CSDH showed rapid resolutions after surgical evacuations. Both of the 2 pathologies were located in the right cranial cavity, compressing the brain parenchyma. Also, serial CTs of the patient did not show any abnormal findings around the medulla oblongata. Furthermore, reported hiccup-associated supratentorial lesions were predominantly located in the right cerebral hemisphere [4,6,8,9,11,14]. Therefore, we assumed that the ICH and CSDH might have caused persistent hiccups by compressing the right cerebral hemisphere. Myocardial infarction/ischemia can be an underlying etiology and a major causative risk factor of cardiovascular hiccups [3]. Although our patient had a medical history of coronary heart disease, neither chest symptoms nor abnormal electrocardiographic findings were identified, mitigating the likelihood of cardiovascular etiology for persistent hiccups. It has been postulated

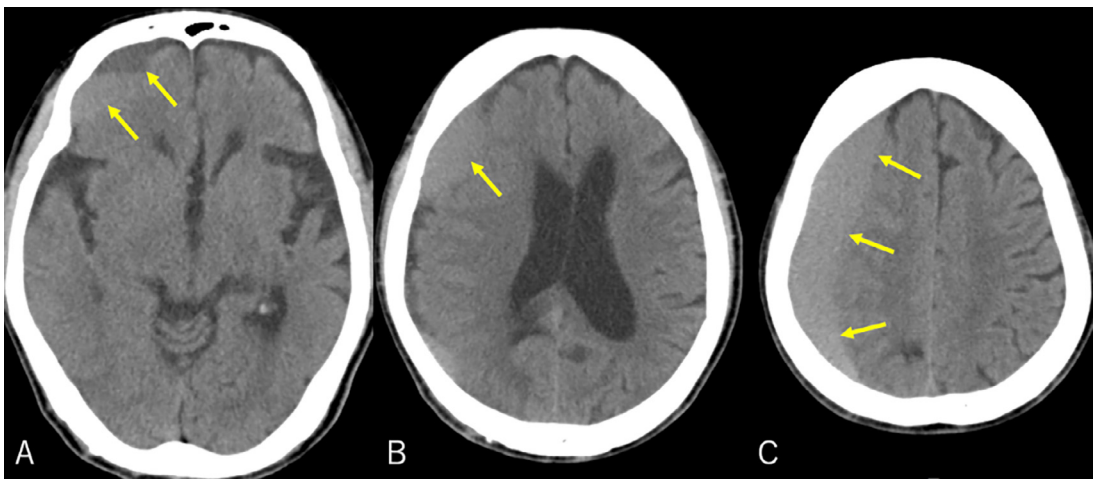

Fig. 3 – Noncontrast axial computed tomography scans at the level of the lower anterior horn (*A*), body of the lateral ventricle (*B*), and centrum semiovale (*C*), performed on the 50th postoperative day, reveal compressive chronic subdural hematoma in the right frontoparietal convexity, measuring 23 mm in maximal thickness. The hematoma mostly appears isodense, with accompanying fluid-to-fluid levels (*arrows*).

that the peripheral phrenic nerve, vagal nerve, sympathetic pathways, and midbrain may be involved in the genesis of hiccups [1,3–7]. In addition, a fraction of supratentorial lesions caused hiccups [4,8-14]. Among them, 2 patients were affected in the right temporal lobe with the remaining one in the right frontal lobe. There 3 showed a remarkable symptomatic resolution after surgical resection [8,9,14]. The cerebral cortices, especially in nondominant hemisphere, may associate with the development of central hiccups.

In the present case, development of CSDH in the right cerebral convexity might be associated with a rapid decompression during ICH evacuation on the ipsilateral hemisphere that can result in a significant displacement of the cerebral cortices, dilation of the subdural spaces, and stretch of the cortical veins, in addition to daily anticoagulant use. To our knowledge, there is no prior documentation implicating CSDH as a cause of hiccups. Recent investigations have posited a correlation between headache and an increased intrahematoma pressure of $\geq$28 cm H_2O [15]. However, our patients did not present with headache or nausea at the time of CSDH diagnosis. Additionally, CT findings did not suggest a significant cerebral compression of the CSDH (Fig. 3). Therefore, we assumed that a mild increase in CSDH pressure might cause persistent hiccups by exerting a gentle pressure to the adjacent cerebral cortices. CSDH may cause persistent hiccups in certain circumstances.

Conclusion

Persistent hiccup warrant consideration as potential manifestation of supratentorial lesions. Hiccups attributed to ICH and CSDH represent a unique clinical entity, with the potential for rapid resolution following surgical evacuation.

Author contributions

All the authors contributed equally to the study.

Ethical standards

We declare that all procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee and the 1964 Declaration of Helsinki and its later amendments.

Patient consent

Written informed consent was obtained from the patient for publication of anonymized data.

REFERENCES

[1] Chang FY, Lu CL. Hiccup: mystery, nature and treatment. J Neurogastroenterol 2012;18(2):123–30.
[2] Misu T, Fujihara K, Nakashima I, Sato S, Itoyama Y. Intractable hiccup and nausea with periaqueductal lesions in neuromyelitis optica. Neurology 2005;65(9):1479–82.
[3] Yuan SM. Myocardial ischemia presenting with hiccups. J Coll Physicians Surg Pak 2019;29(5):469–73.
[4] Itabashi R, Endo K, Saito T, Fukuma K, Yazawa Y. Supratentorial infarcts accompanying hiccups. Brain Behav 2019;9(11):e01439.
[5] Kanbayashi T, Sonoo M. The course of facial corticobulbar tract fibers in the dorsolateral medulla oblongata. BMC Neurol 2021;21(1):214.
[6] Kumral E, Acarer A. Primary medullary haemorrhage with intractable hiccup. J Neurol 1998;245(9):620–9.
[7] Thaci B, Burns JD, Delalle I, Vu T, Davies KG. Intractable hiccups resolved after resection of a cavernous malformation of the medulla oblongata. Clin Neurol Neurosurg 2013;115(10):2247–50.
[8] Hu Z, Cui L, Li W, Liang Z, Du M. Three-year intractable hiccups: compression of the right frontal lobe by meningioma. Am J Med 2017;130(1):e17–18.
[9] Jansen PH, Joosten EM, Vingerhoets HM. Persistent periodic hiccups following brain abscess: a case report. J Neurol Neurosurg Psychiatry 1990;53(1):83–4.
[10] Longatti P, Baseldella L, Moro M, Ciccarino P, Franzini A. Refractory central supratentorial hiccup partially relieved with vagus nerve stimulation. J Neurol Neurosurg Psychiatry 2010;81(7):821–2.
[11] Morita K, Nuki Y, Hashizume H, Togo M. Persistent hiccups induced by supratentorial infarcts and successful treatment with a combination of perampanel and bacrofen: a case report. Clin Neuropharmacol 2022;45(5):135–8.
[12] Shankar T, Pillai A, Kaeley N, Vempalli N. Intractable hiccups as the harbinger of a diffuse pontine glioma. BMJ Case Rep 2022;15(3):e247830.
[13] Stern G. Central stridor from an ependymoma. Mov Disord 1987;2(4):313–16.
[14] van Durme CM, Idema RN, van Guldener C. Two rare complications of glioblastoma multiforme: persistent hiccup and acquired haemophilia A. Neth J Med 2008;66(7):286–8.
[15] Tsutsumi S, Sugiyama N, Ueno H, Ishii H. Are there characteristic indicators for increased pressure in chronic subdural hematoma? J Clin Neurosci 2024;121(3):42–6.