

Case Report

Reversible Hemichorea Caused by Hydrocephalus with Right Sylvian Hematoma Following Subarachnoid Hemorrhage

Yuhei Takado^{1,2*}, Tetsutaro Ozawa², Keiichi Nishimaki³, Jun Maruya³, Hideaki Ishiguro¹, Masatoyo Nishizawa² and Takashi Minakawa³

¹Department of Neurology, Akita Red Cross Hospital, Akita, Japan,

²Department of Neurology, Brain Research Institute, University of Niigata, Niigata, Japan

³Department of Neurosurgery, Akita Red Cross Hospital, Akita, Japan

Corresponding author

Yuhei Takado, Institut de physique des systèmes biologiques, Faculté des sciences de base, Ecole polytechnique fédérale de Lausanne, Switzerland CH-1015 Lausanne, Switzerland, Email: yuhei.takado@epfl.ch

Submitted: 19 August 2013

Accepted: 21 October 2013

Published: 23 October 2013

Copyright

© 2013 Takado et al.

OPEN ACCESS

Abstract

Hydrocephalus has been reported to cause bilateral chorea; however, the relation between hemichorea and hydrocephalus remains unclear. In this study, we describe a patient with hemichorea that worsened in parallel with the progression of hydrocephalus with right sylvian hematoma after subarachnoid hemorrhage (SAH). Within days of ventriculo-peritoneal (VP) shunt placement, the choreatic movements disappeared. Considering that hydrocephalus symmetrically affects the central nervous system, the asymmetric presentation of symptoms in our patient might have been due to the rare underlying pathology, namely, the right sylvian hematoma and the subsequent occurrence of hydrocephalus. Our observation suggests that hydrocephalus can cause hemichorea in the presence of additional factors such as sylvian hematoma, which locally increased pressure on the basal ganglia and surrounding areas. VP shunt placement should be considered as a therapeutic option in chorea patients presenting with hydrocephalus.

INTRODUCTION

Chorea, an involuntary movement disorder characterized by a continuous flow of random, brief, involuntary muscle contractions, originates from dysfunctional neuronal networks interconnecting the basal ganglia and frontal cortical motor areas [1]. Hydrocephalus has been reported to cause bilateral chorea [2-4]. However, the mechanism of hemichorea induced by hydrocephalus remains unclear.

Hydrocephalus is a common and potentially devastating complication of aneurysmal subarachnoid hemorrhage (SAH). Its incidence is approximately 20% to 30%, and its onset can be acute within 48 hours after SAH, or rarely chronic occurring in a delayed fashion weeks and even months after the hemorrhage [5].

In this case report, we present a patient with hemichorea that worsened in parallel with the progression of chronic hydrocephalus with right sylvian hematoma after SAH. However, the previous reports showed that chorea associated with hydrocephalus occurred on both sides without the laterality

of the brain damage [2-4]. To the best of our knowledge, there was only one report that demonstrated the efficacy of VP shunt placement for treating chorea associated with hydrocephalus [4]. Our observation may add new information about the mechanism underlying the chorea induced by hydrocephalus

CASE PRESENTATION

A 77-year-old man presented with a sudden onset of headache and vomiting. He was admitted to our hospital with mild drowsiness and left hemiparesis. Computed tomography, which was performed on the day after symptom onset, showed diffuse SAH with a right sylvian hematoma (Figure 1A). Four-vessel cerebral angiography showed an aneurysm at the M1-M2 bifurcation of the right middle 66 cerebral artery. Evacuation of the hematoma, successful clipping of the aneurysm, and cisternal drainage were performed 3 days after the symptom onset. While the postoperative course was complicated by confusion and left hemiparesis that were caused by cerebral edema around the right sylvian hematoma, those symptoms were gradually improved by glycerol administration within a month after the

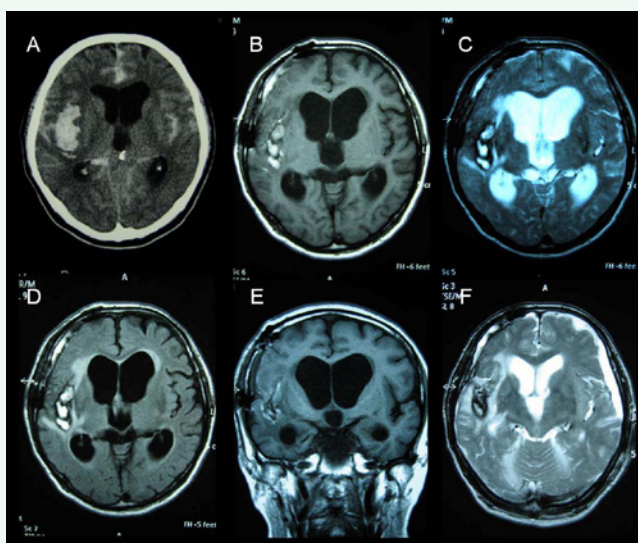


Figure 1 (A): Computed tomography, which was performed on the day after SAH symptom onset, showed diffuse SAH with right sylvian hematoma. (B-E): MRI obtained 42 days after SAH showing hydrocephalus, right sylvian hematoma, and brain edema around the right putamen. (F): Axial T2-weighted MRI obtained 53 days after the SAH onset showing no evidence of hydrocephalus. Hydrocephalus is more obvious in B-E than in A, but the right sylvian hematoma shown in A is bigger than the one shown in B-E. At the time of A, the patient didn't have hemichorea. Hemichorea occurred on the left hand at the time of B-E. The patient was free from hemichorea at the time of F, which were taken 8 days after a VP shunt placement. Cognitive function improved and hemichorea disappeared within a few days after VP shunt placement. (B. axial T1-weighted MRI; C. axial T2-weighted MRI; D. axial fluid-attenuated inversion-recovery MRI; E. coronal T1-weighted MRI).

SAH onset. Thirty days after the SAH onset, the patient developed cognitive impairment and hemichorea that affected the left hand. The choreic movements in the left hand were increased with action, but they were decreased with relaxation. He did not have dopaminergic medication, neuroleptics, or any other drug known to induce chorea [6]. His family history was negative for Huntington's disease or other hereditary choreas.

Cerebral magnetic resonance imaging (MRI) performed 42 days after the SAH onset revealed hydrocephalus and right sylvian hematoma (Figure 1B-E). Mild brain edema was observed around the right basal ganglia, but there were no signs of recent focal lesions in the basal ganglia or subthalamic nucleus (Figure 1B-E). A VP shunt was placed 45 days after the SAH onset. Within a few days after VP shunt placement, cognitive function improved and hemichorea disappeared. Repeat cerebral MRI, which was performed 53 days after the SAH onset, revealed improvement of hydrocephalus (Figure 1F). He had ordinary food during the hospitalization after his recovery from the acute stage of the SAH.

DISCUSSION

Chorea can result from a variety of causes, including infection, autoimmune disease, genetic mutation, neurodegeneration, stroke, neoplasm, drug exposure, and metabolic disease [1]. The patient presented here did not have any of these causative conditions for chorea. There were no recent focal lesions in the basal ganglia at the time of hemichorea, and hydrocephalus with right sylvian hematoma was the only abnormality present.

Although the right basal ganglia was highly vulnerable to brain edema caused by the right sylvian hematoma, which may have caused hemichorea, it is notable that hemichorea worsened in parallel with the progression of hydrocephalus and disappeared shortly after VP shunt placement. In addition, hemichorea presented after the right sylvian hematoma had decreased in size (Figure 1 B-E), rather than when it was at its largest size in the initial stages of SAH (Figure 1F). The occurrence of hydrocephalus might have accentuated the harmful effect of the right sylvian hematoma, which locally increased pressure on the right basal ganglia and surrounding areas, leading to dysfunction of basal ganglia connectivity. Our observations are in accordance with the notion that dysfunction of basal ganglia connectivity to the surrounding area can cause chorea [7].

There are only a few reports (four patients in three reports) of hydrocephalus-associated chorea [2-4], and all four of these patients presented with bilateral chorea. Three of the four patients had Huntington's disease as well as hydrocephalus, and the bilateral chorea of these patients moderately responded to treatment of the hydrocephalus [2,3]. The remaining patient had brainstem tumor as well as the bilateral chorea associated with hydrocephalus, which was successfully treated by VP shunt placement [4]. Notably, there was another case report showing that brainstem tumor itself caused hemichorea [8]. Although chorea associated with hydrocephalus is very rare, these observations suggest that hydrocephalus may facilitate the development of chorea or hemichorea that is originated from underlying diseases. These observations also support the concept that the occurrence of hydrocephalus in our patient might have accentuated the harmful effect of the right sylvian hematoma, which locally increased pressure on the right basal ganglia and surrounding areas, leading to the manifestation of hemichorea. Taken together, our observations suggest that VP shunt replacement may be helpful in treating chorea associated with hydrocephalus.

CONCLUSION

Considering that hydrocephalus symmetrically affects the central nervous system, the asymmetric presentation of symptoms in our patient might have been due to the rare underlying pathology, namely, the right sylvian hematoma and the subsequent occurrence of hydrocephalus. This report adds new information that hydrocephalus can cause hemichorea in the presence of additional factors such as sylvian hematoma, which locally increases pressure on the basal ganglia and surrounding areas. Therefore, VP shunt placement should be considered for this condition. Physicians should be aware of this rare case of treatable hemichorea associated with hydrocephalus.

REFERENCES

1. Francisco C, Klaus S, Katherina JM, Gregor KW, Werner P. Seminar on choreas. *Lancet. Neurol.* 2006; 5: 589-602.
2. Shirani P, Salamone AR, Lahijani E, York MK, Schulz PE. Lumbar puncture alleviates chorea in a patient with Huntington's disease and normal pressure hydrocephalus. *Behav Neurol.* 2009; 21: 193-195.
3. Tang BH, Lieberman A, Rovit R. Huntington's chorea associated with normal pressure hydrocephalus. *Eur Neurol.* 1975; 13: 189-194.
4. Voermans NC, Schutte PJ, Bloem BR. Hydrocephalus induced chorea. *J*

- Neurol Neurosurg Psychiatry. 2007; 78: 1284-1285.
5. Germanwala AV, Huang J, Tamargo RJ. Hydrocephalus after aneurysmal subarachnoid hemorrhage. *Neurosurg Clin N Am.* 2010; 21: 263-270.
 6. Bhidayasiri R, Truong DD. Chorea and related disorders. *Postgrad Med J.* 2004; 80: 527-534.
 7. Postuma RB, Lang AE. Hemiballism: revisiting a classic disorder. *Lancet Neurol.* 2003; 2: 661-668.
 8. Patankar AP. Hemi-chorea: an unusual presentation of brainstem glioma. *Br J Neurosurg.* 2013; 27: 256-258.

Cite this article

Takado Y, Ozawa T, Nishimaki K, Maruya J, Ishiguro H, et al. (2013) Reversible Hemichorea Caused by Hydrocephalus with Right Sylvian Hematoma Following Subarachnoid Hemorrhage. *J Neurol Transl Neurosci* 1(3): 1024.