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Chorea accompanied with chronic subdural hematoma: A case report and literature review

Akira Tempaku

ABSTRACT

Introduction: Chronic subdural hematoma (CSDH) presents hemipalsy, gait disturbance, dysarthria, cognitive disorder, or conscious level down. These hypokinetic movement dysfunctions are brought from the downregulation of the cerebral neural activities, which are caused by the hematoma compression. In contrast, a rare case of CSDH patients shows hyperkinetic movement disorders.

Case Report: An 82-year-old woman presented with chorea in the head and extremities, which was dominant in the right hand. The involuntary movement progressed for last three days. Her conscious level was clear and the function of the lower cranial nerves was normal. She had no hemipalsy nor sensory disorders. Brain magnetic resonance image revealed a chronic subdural hematoma in left hemisphere. After the removal of hematoma through surgical irrigation and evacuation, the chorea disappeared immediately.

Conclusion: Subdural hematoma compressed cerebral cortex, which disturbed the neural network function. Especially, modulation of the cortico-basal ganglia loop function by subdural hematoma causes to the involuntary movement disorders as chorea. This report describes a rare case of chorea associated with CSDH. Previous case literatures are reviewed and summarized.

Keywords: Chorea, Chronic subdural hematoma, Cortico-basal ganglia loop

Akira Tempaku[1]

Affiliation: [1]MD, Director, Department of Neurosurgery, Hokuto Hospital, Obihiro, Hokkaido, Japan.

Corresponding Author: Akira Tempaku, Department of Neurosurgery, Hokuto Hospital, 7-5 Inada-cho-kisen, Obihiro, Hokkaido, Japan.

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INTRODUCTION

Chorea is brought to the dysregulation of the cortico-basal ganglia loop function [1–3]. Huntington's disease presents chorea by their neurodegenerative result [4–6]. Post-stroke state, malignancy in brain, metabolic diseases, or infectious diseases also cause to the involuntary movements [5, 6]. Although cerebral deformity by intracranial hematoma can modulate central nerve system function, subdural hematoma has rarely focused as a pathogenesis of the hyperkinetic movement disorder including chorea. Here reports a rare case of chorea accompanied with chronic subdural hematoma (CSDH).

CASE REPORT

An 82-year-old woman presented with chorea like movement disorder, which was right dominantly. She had no medication of cholinergic or dopaminergic agents. Head computed tomography (CT) showed the left CSDH (Figure 1). No obvious lesion of stroke and tumor were observed in her brain by magnetic resonance imaging (data not shown). Metabolic disorders or infectious signs in central nerve system were excluded by serum test. Left brain was mildly compressed by subdural hematoma. Hemipalsy or verbal disturbance was not obvious. Basal ganglia and surrounding white matter were packed slightly tight. Burr hole surgery with irrigation and evacuation of hematoma was performed.

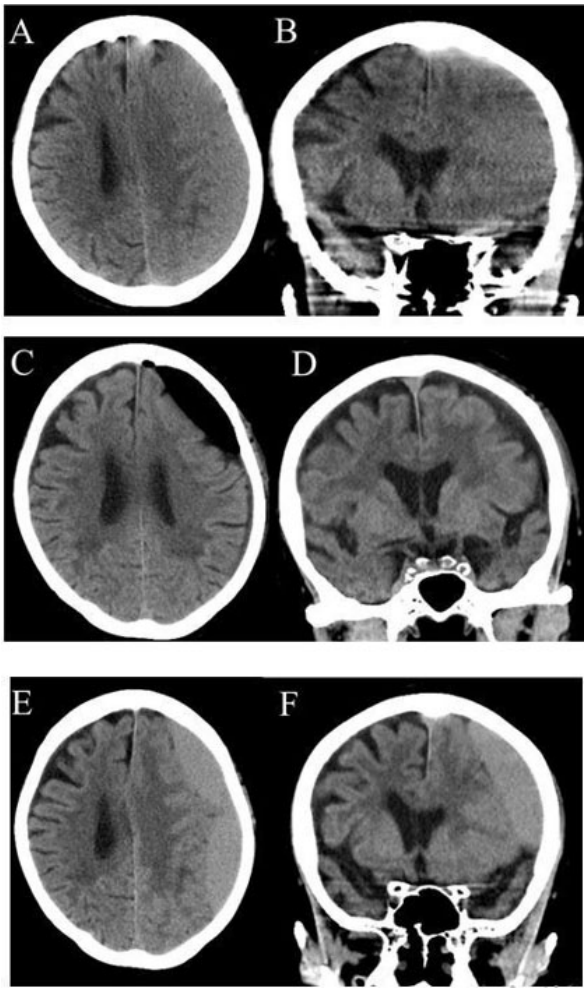

Figure 1: Head computed tomography (CT) was performed at the first time admission (A, B) and after the first time irrigation (C, D). Head CT was performed at the second time admission with chorea recurrence (E, F). A, C, and E show head CT with axial view. B, D, and F show head CT with coronal view.

After the cerebral compression was relieved, the chorea disappeared. Three weeks later, subdural hematoma restored with chorea recurrence. Second time irrigation was performed and chorea vanished in immediate time. Because of no subdural hematoma was observed in the post-second time surgical operation days, chorea was never seen in her extremities.

DISCUSSION

Left chronic subdural hematoma was accompanied with chorea as right upper limb dominant manner. Hyperkinetic movement disorder disappeared immediately after the hematoma evacuation. Although the gradual recurrence of chorea was observed following to the subdural hematoma reformation, re-evacuation of hematoma vanished chorea promptly. These medical treatments course demonstrated that chorea was brought from the compression of cerebral hemisphere.

Chorea is known to be classified into primary ones and secondary ones. Hereditary chore is associated with Huntington's disease [4, 5, 7] or Dentato Rubro Pallido Lysine Atrophy (DRPLA) [8, 9], which are based on the amplified cytosine-adenine-guanine (CAG) repeat in the Huntington gene or DRPLA gene. These heredity diseases have family history of chorea because of their autosomal dominant hereditary manner. Since this patient case had no family history about the movement disorders, the hereditary chorea was denied. Benign hereditary chorea [10] and paroxysmal kinesigenic choreoathetosis [11], which mainly present infancy, were excluded from the diagnosis.

Secondary chorea is caused by various etiologies [5, 6, 12]. Inflammatory diseases including Sydenham chorea with Group A Streptococcus infection [13] was denied by her no history of previous infection. Toxic induction from nitric oxide, alcohol, diphenylhydantoin, carbamazepine, L-dopa, or phenothiazine [5, 6] was not likely to the etiologies since no medical history. Metabolic disturbances including Wilson's disease, Fahr disease, Leigh disease, Loius–Barr syndrome, Lesch–Nyhan syndrome, Hallervorden–Spatz disease, Pelizaeus–Merzbacher disease, lipidosis, mucopolysaccharidosis, or acanthocytosis [5, 6] were excluded with her other clinical symptoms or serological tests. Brain tumors at the basal ganglia or brain stem were not detected by cerebral MRI at the admission time. Cerebral vascular anomalies including arterial-venous shunt and arterial malformation were not observed in brain MRI. Hemorrhagic or ischemic change of lenticulostriate artery perfusion area, thalamus, or mid brain did not diagnose, either. Only hematoma formation of subdural space and cerebral parenchymal compression were observed. Although the obvious head injury history was not obtained, chronic subdural hematoma might be brought by the head trauma. Decompression of cerebral hemisphere by hematoma irrigation and evacuation resulted in the chorea disappearance. These treatment history also revealed that chorea was brought from the compression of cerebral parenchymal by CSDH.

The pathogenesis of chorea might be caused by disturbances in the cortico-basal ganglia loop function [6] (Figure 2A). Degeneration of caudate neuron, which mediates with γ-aminobutyric acid (GABA), causes the decrement of suppressive signal against the globus pallidus externalis (GPe) segment. Then disinhibited GPe excites to give the suppressive signal against hypothalamus via GABA neuron. Suppressed hypothalamus dysregulates the enhancement of globus pallidus internals (GPi)/substantia nigra reticular formation (SNr) activities, which inhibits thalamic activities via GABA neuron. Increased thalamic signal, which potentially upregulates cerebral cortical activities and enhances to the spine cord signals, results in hyperkinetic movement disorders [3, 14, 15] (Figure 2B).

Present case has been explained that the compression of cortical parenchymal by CSDH caused to damage

caudate, which brought the modulation of the cortico-basal ganglia loop activities. Then, decreased indirect pathway signal, which inhibited the hyperkinetic involuntary movement, resulted in the chorea-like movement disorder. Previous report also mentioned and summarized chorea patients associated with CSDH [13] (Table 1). Because of hematoma removal vanished chorea, it was concluded that cerebral compression caused chorea. However, some cases showed the ipsilateral or generalized chorea. Even if the secondary chorea is brought from the cerebral dysfunction by hematoma compression, only contralateral chorea is observed. Ipsilateral chorea might be obtained from the tight hematoma compression, which compressed the contralateral caudate and modulated its function. Available head CT or MRI in previous reports and the presented case figure have suggested that chorea had usually been brought by the hematoma, which predominantly affected the frontal and lateral portions. These hematomas tend to compress the frontal lobe rather than temporal or parietal lobe. Frontal lobe compression from front temporal side usually extends toward the caudate position anatomically. Furthermore, available head images have described that the shift and narrowing of frontal horn have been massive than that of posterior or inferior horn. These image findings would suggest to the etiology of chorea. In addition, cases of tightly shifted frontal falx might be brought the ipsilateral chorea by contralateral caudate compression.

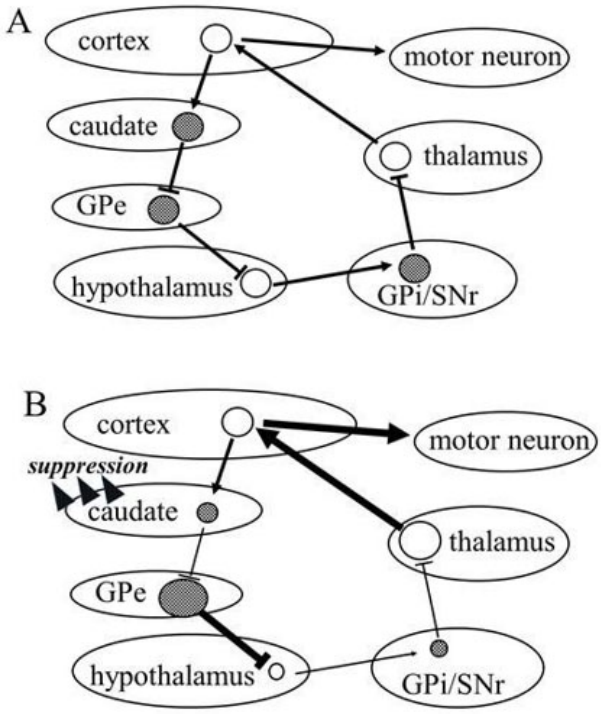

Figure 2: Schematic depiction of the cortico-basal ganglia loop function. (A) Illustration of normal regulation of basal ganglia. (B) Modulated neural network with chorea is shown. White circle means the glutamate acid mediating neuron, which enhances the secondary neuron activity. Plain circle means the γ-aminobutyric acid (GABA) mediating neuron, which inhibits the secondary neuron activity. Circle size visualizes the neuron excitement level. While large circle means enhanced action potential, small circle means inhibited action potential. Arrow means the glutamate neuron signal. Resistant arrow means the GABA neuron signal. Thickness of arrow and resistant arrow associates to the intensity of neural signal. While increased signal is shown by thick line, decreased signal is shown by thin line.

CONCLUSION

This case report describes a rare case of chorea associated with CSDH. Hematoma compression to cerebral parenchymal might cause to downregulate caudate activities. Chorea was obtained by the result of the cortico-basal ganglia loop modulation. Unusual clinical feature, which shows the hyperkinetic movement disorder, is also kept in mind to diagnose CSDH. Especially, the hematoma, dominant in front temporal side with massive compression of caudate and frontal horn, might cause to the chorea.

Table 1: Cases summary of chorea associated with chronic subdural hematoma

No.	Author (year)	Age/sex	Side of hematoma	Side of chorea	Outcome of hematoma removal	Associated factors
1	Bae et al. (1977) [16]	12/male	Left	Generalized chorea	Immediate disappearance	Methotrexate administration
2	Gilmore (1979)	64/male	Right	Left hemichorea	No surgery	–
3	Vincent (1980) [17]	83/male	Right	Right hemichorea	1 week continuance	No family history; no MRI study
4	Bae et al. (1980) [18]	57/female	Bilateral	Generalized chorea	4 days continuance	Not mentioned
5	Bae et al. (1980) [18]	47/female	Bilateral	Generalized chorea	Immediate disappearance	Family history of Huntington's disease
6	Kotagal et al. (1981) [19]	73/male	Bilateral	Generalized chorea	Disappearance	Not mentioned
7	Saito et al. (1982) [20]	52/male	Right	Generalized chorea	Immediate disappearance	No family history; no MRI study

Table 1: *(Continued)*

No.	Author (year)	Age/sex	Side of hematoma	Side of chorea	Outcome of hematoma removal	Associated factors
8	Matsumoto (1986) [21]	67/male	Left	Right hemichorea	No surgery	Not mentioned
9	Yoshikawa et al. (1992) [22]	73/male	Left	Left hemichorea	1 month continuance	Stenosis of right middle cerebral artery
10	Sung et al. (2004) [23]	76/female	Bilateral	Generalized chorea	2 days continuance	Flunarizine administration
11	Kobal et al. (2007) [24]	71/male	Bilateral	Generalized chorea	A few hours continuance recurrence at 2 days	41 CAG repeat on IT15 gene
12	Young et al. (2008) [25]	80/female	Left	Generalized chorea	48 hours continuance	Not mentioned
13	Pistacchi et al. (2016) [26]	76/female	Right	Generalized chorea	A few days continuance	Not mentioned
14	Shiraishi et al. (2018) [13]	86/female	Left	Generalized chorea	1 week continuance	Amantadine administration
15	Present case (2024)	82/female	Left	Right hemichorea	Immediate disappearance	No family history; no cerebral lesions in MRI

The table is referred and modified based on Shiraishi et al.'s article [13].

Abbreviation: MRI: magnetic resonance image.

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Author Contributions
Akira Tempaku – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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Author declares no conflict of interest.

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All relevant data are within the paper and its Supporting Information files.