



Resolution of Stroke-Related Hemichorea-Hemiballismus with Haloperidol

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Hemichorea-hemiballismus (HC-HB) is a hyperkinetic disorder characterized by violent, unilateral jerking movements which usually improve to choreoathetosis with time. Etiology of HC-HB has been demonstrated in case reports and series, with acute stroke being one of the known etiologies. Among the rare post-stroke hyperkinetic syndromes, HC-HB is the most common. However, there is limited data on management and long term follow up of this debilitating condition when related to acute stroke. Here we describe a 79-year-old gentleman with acute caudate stroke, with resulting HC-HB, whose symptoms were managed with haloperidol while hospitalized. Resolution of HC-HB and subsequent discontinuation of haloperidol yielded an optimal patient outcome. Long term follow-up for this patient has demonstrated lack of symptom recurrence at 1 year. This case further supports the use of neuroleptics such as haloperidol as first line for management of HC-HB.

Keywords: Stroke; Hyperkinesias; Dyskinesia; Haloperidol

INTRODUCTION

Acute hemichorea-hemiballismus (HC-HB) is a consequence of approximately < 1% of all stroke^{1,2}. Hemiballismus is a flinging, high amplitude and commonly violent movement of proximal limbs. Hemichorea is characterized by continuous, involuntary irregular movements, usually of slower cadence and lower amplitude.

Disruption of dopamine regulation may be the mechanism by which stroke originating within the basal ganglia can elicit HC-HB³. Management of these acute symptoms, therefore, have typically included dopamine antagonism, with variable success.

Here we detail a case of an elderly gentleman with an ischemic stroke, presenting as fluctuating aphasia and dysarthria that re-

solved and was followed development of persistent HC-HB within 24hrs of last known well. Management with haloperidol for his striking symptoms demonstrated significant improvement, allowing for discontinuation of the agent, without symptom recurrence by 1 year post stroke.

CASE

A 79-year-old gentleman with vascular risk factors of hypertension, hyperlipidemia, pre-diabetes, obesity, psoriasis and prior nicotine use was brought in by his wife to a primary stroke center after awakening with aphasia and mild dysarthria. He presented five hours from his last known well (LKW). Local stroke protocol was initiated, and noncontrast head computed tomography (CT) scan

noted left sided hyperdense MCA (middle cerebral artery) sign, consistent with acute thrombus. This was confirmed as a non-occlusive left M2 MCA thrombus by CT angiography of head and neck (Fig. 1A). Clinically, his National Institutes of Health Stroke Scale (NIHSS) was 0 and he had complete resolution of dysarthria thus intravenous (IV) thrombolysis (rtPA) or endovascular stroke treatment (EVT) were not indicated. He was given dual antiplatelet therapy (DAPT) and admitted to the hospital under observation.

While in the ED, his expressive aphasia briefly recurred, again spontaneously resolving 15 minutes later. Due to this stuttering course, he was transferred to a comprehensive stroke center.

During transfer to our comprehensive stroke center, he was noted to develop severe “right sided chorea-type movements” involving the arm, leg, and – to a lesser degree – face. Formal NIHSS remained 0, and he was still not considered to be a candidate for EVT. A trial of Ativan was successful in achieving an adequate MRI quality image, revealing infarctions to the caudate head, insular cortex, and corona radiata (Fig. 1B). Magnetic resonance angiography showed interval resolution of the thrombus involving the superior division of the left M2 MCA, but persistent occlusion of the inferior division. That evening, nursing noted worsening hyperkinetic movements such that he was unable to sleep due to violent right sided movements of his arm and leg and concern for resultant bruising of the right arm.

On hospital day (HD) 1, right sided movements persisted, developing more choreiform character and was noted to include his face. His wife also noted hyperverbosity, which was atypical for him. Decision was made to attempt to palliate symptoms with oral

haloperidol 1 milligram twice daily. After one dose the patient experienced a decrease in the severity of symptoms, without negative sedating side effects and decision was made to increase dose to 2mg BID starting that evening. By that night movements were persistent however improving in the leg and face and he was able to sleep.

HD 2 was notable for significant improvement in all right sided movement by his third dose of haloperidol. By HD 3, his movements were minimal and his speech prosody returned to normal. Continued improvement in symptoms lead to haloperidol discontinuation by HD 4.

Stroke etiology was determined to be cardioembolic as workup as continuous telemetry revealed paroxysmal atrial fibrillation. Further telemetry monitoring demonstrated tachybradycardia syndrome and dual chamber pacemaker was placed by cardiology. DAPT was discontinued and direct oral anticoagulation (DOAC) was initiated the day after surgical procedure, on HD 4. After initial recommendations to discharge to a skilled nursing facility, his motor symptoms improved such that he was discharged home on HD 8. By time of discharge, no HC-HB movements were noted. By 1 year of follow up the patient has not had recurrence of symptoms.

DISCUSSION

Acute HC-HB is a rare consequence of stroke, with management generally accepted to involve dopamine antagonism, with variable results^{2,4}. Here, we discuss a patient presentation of stroke with stuttering onset, likely of cardioembolic origin, with involvement of the caudate, insular cortex, and corona radiata. Develop-

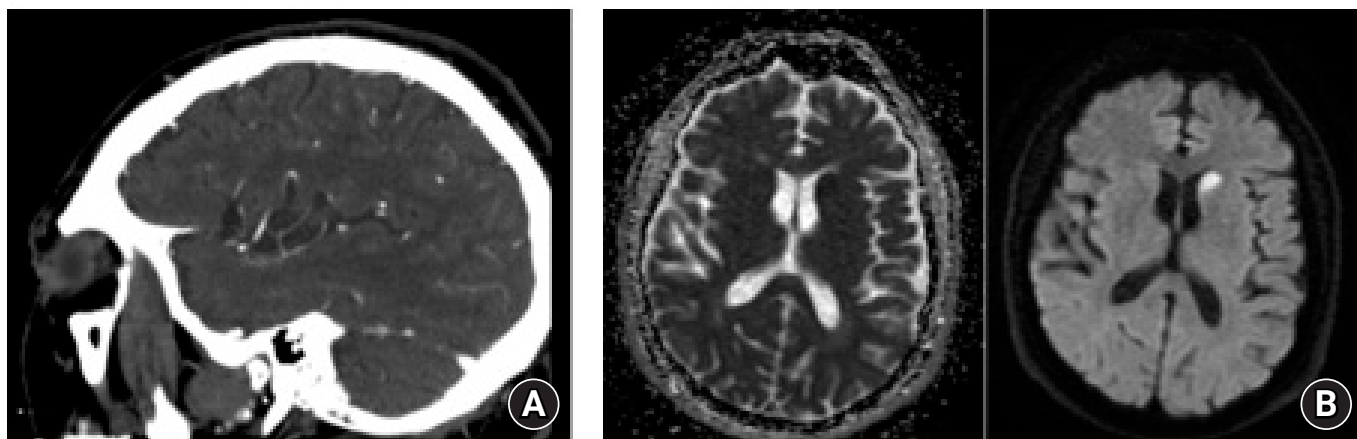


Fig. 1. (A) Computed tomography imaging from primary stroke center, demonstrating M2 lesion. (B) MRI images (left: apparent diffusion coefficient and right: diffusion weighted image) at comprehensive stroke center, after onset of hemichorea-hemiballismus symptoms. MRI: Magnetic resonance imaging.

ment of HC-HB was noted within the first 24 hours and was successfully palliated with a trial of haloperidol.

While HC-HB is an overall rare symptom of stroke, amongst the post-stroke hyperkinetic symptoms, it is the most common. Risk factors for development of this phenomenon include location of stroke, as well as older age and possibly female gender^{5,6}.

Other conditions such as hyperglycemia (coined diabetic striatopathy)^{7,8}, less commonly vascular malformations, CNS toxoplasmosis in AIDS patients⁴, tuberculoma⁹, and amphetamine use¹⁰ (although usually bilateral motor symptoms) have been published as alternative etiologies for this phenomenon.

Neuroanatomical classic teaching detail that insults to the contralateral subthalamic nucleus (STN) result in hemiballismus. More recently, a review of 29 patients with stroke which produced HC-HB were collated onto a reference brain map, identifying a broader network that may underlie this clinical phenomenon. Culprit lesions included: STN, caudate, putamen, other subcortical white matter, and cortex with each case demonstrating connectivity to the posterolateral putamen³.

It is postulated that the underlying pathophysiology of HC-HB involves dysfunction in the Direct and Indirect Extrapyramidal pathway. This pathway functions to modulate the activity of cortical motor neurons thus facilitating voluntary movements and suppressing any unwanted involuntary movements. Striatal lesions result in increased excitation of motor cortex due to loss of thalamic inhibition resulting in unopposed motor activity and unwanted movements such as HC-HB.

Symptom onset has been documented as variable, with most occurring within 24 hours, however also commonly within one week of stroke onset⁶. It is typical, if present, for ballistic movements to occur at onset of symptoms, with a transition to choreiform movements, and sometimes dystonia. All aspects of the symptomatology, evolution, and timing (within 12 hours from initial symptom) were fitting in our patient. We hypothesize the initial presentation with aphasia and dysarthria to be more reflective of hypoperfusion in the superior M2 territory, with resolution of those symptoms upon spontaneous recanalization of the same vessel. The persistent occlusion of the inferior division and lenticulostriate arteries resulting in caudate head DWI restriction was felt responsible for HC-HB in our patient.

Neuroleptics have been used for dopamine antagonism effect, as well as topiramate and benzodiazepines⁴. Tetrabenazine has also been used in a dopamine-depleting manner to reportedly good effect. Dopamine receptor blockers, including first- and second-generation antipsychotics, have been generally considered the most effective agents to reduce the severity of choreiform movements, regardless of the cause. Benzodiazepines may also have a mild an-

ti-chorea effect by potentiating the inhibitory effects of GABA, although the use of such agents is poorly documented. In our case, lorazepam produced useful for mild, temporary symptom improvement in preparation for MRI.

Literature on symptom resolution is variable, with cited recovery in anywhere from 10%–67% of cases^{5,6}. There is a notable number of cases with unknown long term outcomes⁶. It has been suggested that those with a cortical lesion have improved rates of recovery vs. subcortical lesions, with sparse literature to support this.

In our patient, dopamine antagonism with haloperidol yielded remarkable improvement in symptoms, such that the agent itself was able to be discontinued after 4 days of use. Outpatient follow up demonstrated lack symptom recurrence since the initial stroke. This trial of haloperidol, with a meaningful follow up period of 1 year may suggest that, when effective, patients may be able to discontinue their treatment in the post-stroke setting.

CONCLUSION

This case highlights haloperidol as a viable management strategy in a striking presentation of stroke-related HC-HB, with long term follow up demonstrating sustained resolution of symptoms. While the symptom improvement is most notable, other aspects to this patient's presentation, which include caudate head involvement, time of onset, evolution of the motor syndrome, and long-term outcome, add to the literature characterizing this rare, stroke-related phenomenon.

NOTES

Ethics statement

Informed consent was obtained from the patient herein for publication of his clinical information.

Author contributions

Conceptualization: MA, Supervision: BS. Writing – original draft, Writing – review & editing: All authors.

Conflict of interest

There is no conflict of interest to disclose.

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Data availability

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