A rare case of movement disorder in Intensive Care Unit

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Abstract

Hemichorea-hemiballismus syndrome (HCHB) represents a peculiar form of hyperkinetic movement disorder with varying degrees of chorea and/or ballistic movements on one side of the body. The patients are conscious of their environment but unable to control the movements. HCHB is a rare occurrence in acute stroke patients. Patients with sub-cortical strokes are more prone to develop movement disorders than with cortical stroke. We report one such interesting case here posing difficulties in management and intensive care of the patient. The patient remained refractory to all the drugs described in literature, and adequate control of the hyperkinetic movements could be achieved only with continuous intravenous sedation.

Keywords: Dilapidating, hemichorea-hemiballismus, movement disorder, refractory

Introduction

Movement disorders are well recognized in poststroke patients. Varying types of movement disorders both hypo and hyperkinetic has been described. They can manifest at the time of stroke or appear later. Recognition of such disorder is not only important in terms of management but also counseling the family regarding the nature of such movements, how long they are going to stay and how difficult it can get to control them. Hemichorea-hemiballismus (HCHB) movements may resolve spontaneously but sometimes require aggressive measures. Hemiballismus-hemichorea is the part of the same spectrum of involuntary movements, as evident by the term HCHB syndrome.[1,2]

Case Report

A 73-year-old male, hypertensive with no other comorbidities presented with abrupt onset of continuous involuntary movements of the left upper and lower limb for 1 day. These were not associated with limb weakness, slurring of speech or drowsiness. He had accelerated hypertension on admission for which labetalol infusion was started. Central nervous system examination revealed involuntary large amplitude rapid flinging ballistic movements involving left upper and lower limbs. The movements were strictly unilateral, involved proximal parts of limbs more than the distal ones, and had a less prominent choreiform component as well. There was neither cranial or focal motor deficit nor there were pyramidal signs.

All routine investigations were normal. Magnetic resonance imaging brain showed acute hemorrhage in the right thalamic region [Figure 1], along with hypertensive microangiopathic changes. Hence, he was diagnosed with HCHB syndrome secondary to hypertensive thalamic bleed. For control of the

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continuous movements, the patient was initially started on oral tetrabenazine and parenteral haloperidol. The doses were gradually escalated, but the movements could not be controlled. Later on, atypical antipsychotics including quetiapine and clozapine were also tried. In addition to tetrabenazine and a neuroleptic agent, various other drugs were used as an add-on, including clonazepam and antiepileptic drugs such as sodium valproate, levetiracetam, and topiramate.[3] However, the patient remained refractory to oral agents for initial few weeks and had to be kept on infusions of midazolam or dexmedetomidine for an adequate control of the involuntary movements. He required elective intubation and ventilatory support in view of drug-induced sedation. Repeat computed tomography head was done which showed slight resolution of right thalamic bleed. The patient was tracheotomized and weaned off ventilatory support as his requirement for sedatives decreased. The surgical options of stereotactic thalamotomy or deep brain stimulation of thalamus were also discussed with family, but they opted to continue with medical management only. Gradually his condition improved over the weeks, and the movements were adequately controlled on quetiapine, tetrabenazine, and topiramate. The ballistic movements decreased over few weeks, and choreiform movements became the predominant component during the later part of hospital stay. Furthermore, parakinesias (patient’s attempts to incorporate the choreiform movements into semi-voluntary movements) were quite apparent at this stage. He was decannulated and discharged in a stable condition.

Discussion

Hemiballismus is a severe, violent, arrhythmic, and large amplitude movement of a limb from a proximal joint with an element of rotation while hemichorea is a unilateral, rapid involuntary motions of flexion and extension, rotation or crossing, which may involve any body parts, but predominantly distal parts.[1] The most common finding in vascular hemiballismus is a hemorrhagic lesion of the contralateral subthalamic nucleus. Case reports with lesions outside subthalamic area have been described in literature.

The frequency of developing poststroke movements is unclear. In one study, 3.7% of 1500 stroke patients developed movement disorders.[2] In another study of 2500 first stroke patients found that 1% developed an acute or delayed movement disorder.[4] The incidence of abnormal movements poststroke is equal in men and women. A study showed the average age of 63.3 years.[2] The time over which a movement disorder develops is quite variable ranging from the day of onset to several years after the stroke. Basal ganglia are the most common area implicated in poststroke movement disorders. The basal ganglia circuit acts as a cortical feedback loop in which signals from the neocortex are relayed through the striatum, pallidum, and thalamus back to the cortex. The net effect of cortical activity of thalamic outputs is excitatory over the cortex. These direct pathways are modulated by other loops from the substantia nigra (dopaminergic) and subthalamic nucleus. Any interruption of these pathways by focal lesions may lead to movement disorders.[5]

Patients who are destined to develop abnormal movements after a stroke usually do so at the time of presentation. Motor deficit/dysfunction is more pronounced. Sensory involvement is variably described in literature. The Severe sensory loss has been described while others have reported that sensory deficit is rarely present.[6-8]

Although these conditions tend to resolve spontaneously but they do require treatment in view of the distress caused to the patients. Pharmacotherapy with typical and atypical neuroleptics and dopamine-depleting agents is usually given.[9] Typical neuroleptic agents including haloperidol and chlorpromazine are among the first-line drug treatments for hemiballismus. However, the atypical neuroleptic agents such as olanzapine and quetiapine are preferred now in view of lesser risk of tardive dyskinesias with long-term antidopaminergic therapy. Clozapine has also been used in refractory cases. Dopamine depletors like tetrabenazine is also very effective in treating hemiballismus but can cause parkinsonism as well as depression or hypotension. Other drugs which have been used with some success include benzodiazepines, sodium valproate, and...
newer antiepileptic agents. Surgical intervention with deep brain stimulation (DBS) should be considered in drug-resistant cases. Both stereotactic ventral intermediate thalamotomy and chronic thalamic stimulation have been effective. In one case of HCHB secondary to hyperglycemia, DBS reduced chorea and ballism.

Poststroke movements can resolve in few days or can take months to resolve despite medical treatment. In one series, 56% of cases resolved within 40 days, other cases had chorea lasting as long as 41 months. In other study, at 1-year follow-up, 10% of patients with chorea had improved completely and 75% partially.

Our patient had a prolonged intensive care unit/high dependency unit stay of 70 days; finally, he was discharged in a stable condition. He was alert, taking orally and was ambulatory with support at the time of discharge. He is fully independent in all activities after a 1 year follow-up. We had to use very high doses of all the drugs described in literature, in varying combinations, in an attempt to control the HCHB in our patient.

**Conclusion**

HBHC syndrome occurs uncommonly in association with stroke and tends to resolve over time. A variety of pharmacological and stereotactic neurosurgical procedures are available to treat the condition, but the disorder is often refractory to medical management in the acute and subacute stage, and the patient can be salvaged only with a meticulous intensive care.

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**Conflicts of interest**

There are no conflicts of interest.

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